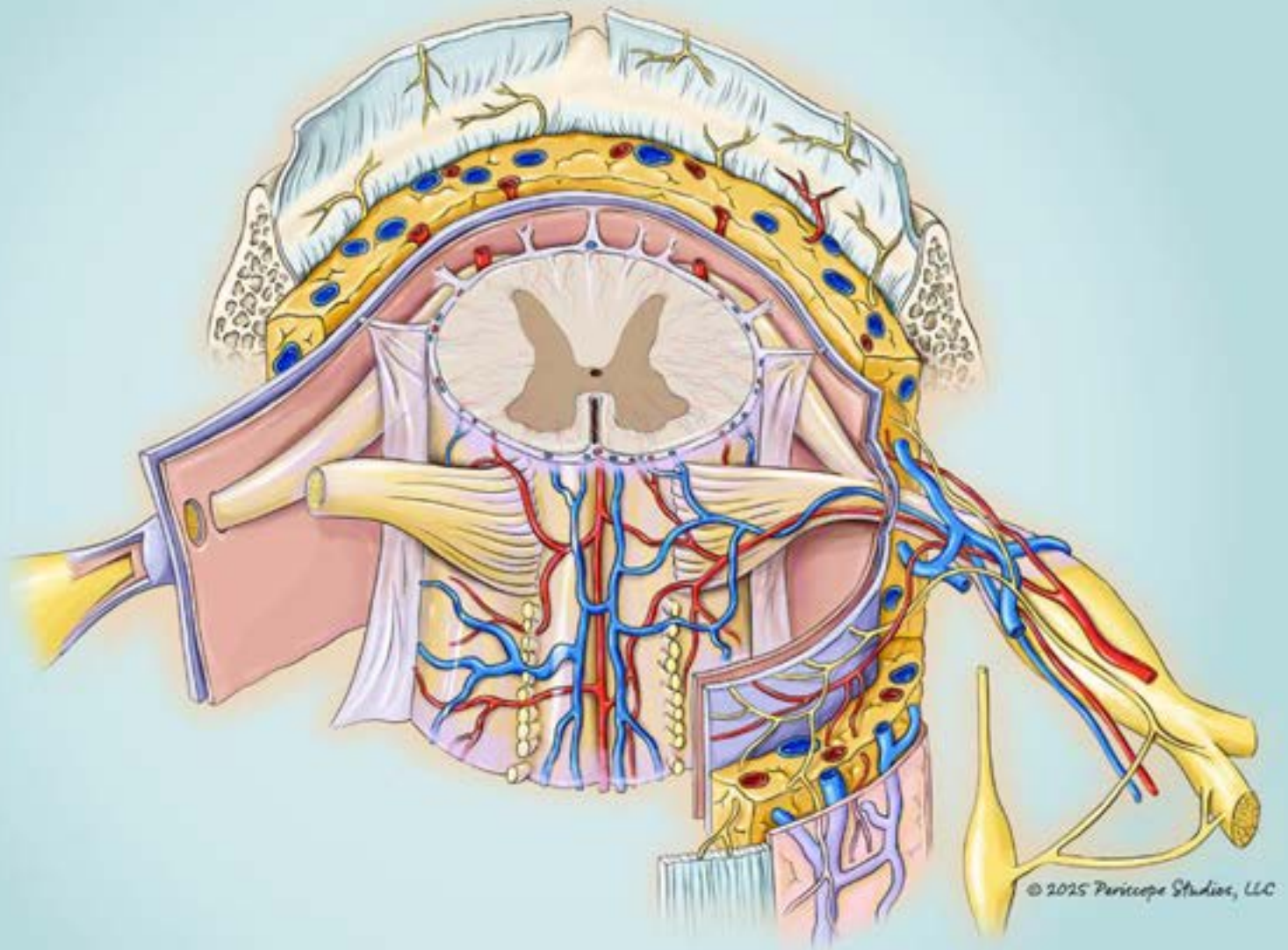


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JBNC

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Assist in strategic planning and development of the journal by evaluating and recommending best practices, organizing and suggesting calls for special or thematic issues, while ensuring that the journal's direction aligns with current trends and needs in the field.

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Submissions must follow the limits and specific requirements outlined on the table below, according to their section.

Section	Abstract	Text sections	Text length	Tables & Figures	References (max)
Original article	structured not exceeding 200 word	introduction, methods, results, discussion, conclusion and references	4000 words	10	75
Review (preferably systematic review)	non-structured not exceeding 200 words	introduction, method, results, discussion, conclusion and references	4000 words	10	75
Case Report (preferably with systematic rev <<	non-structured not exceeding 200 words	introduction (with brief literature review), clinical case presentation, discussion, final comments and references	3500 words	8	45
Brief Note	non-structured not exceeding 200 words	no requirement	1500 words	3	30
Clinical Images	non-structured not exceeding 200 words	no requirement	1500 words	5	30

INTRODUCTION

In the Introduction section we state the motivation for the work presented in the manuscript. Its contents could be: 1) context (to orient readers who are less familiar with the topic and to establish the importance of the manuscript), 2) need (to state the need for the work, as an opposition between what the scientific community currently has and what it wants), 3) task (to indicate what was done in the effort to address the need), and 4) object of the document (to prepare the readers for its structure).

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Patient's clinical data in comprehensive account of the presenting features, with medical, and social, family history, if needed are presented. All crucial investigations to the management of decisions should be discussed. Images of

the case: Choose appropriate images being aware of removing any detail that can identify the patient. If relevant, describe the treatment or surgery. Outcomes and follow-up are described elsewhere.

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In Materials and Methods section, the technical specifications and quantities and source or method of preparation are described. Attention to the use only scientific names of drugs; inclusion of the manufacturer in brackets when describing equipment. Discuss statistical methods if needed.

RESULTS

In Results section, the results of the paper are presented in logical order, using tables and graphs as necessary. Remember that results must be presented and then explained. The results are explained showing how they help to answer the research questions (already cited in the Introduction section).

DISCUSSION

In Discussion section, the principles, relationships and generalizations shown by the results are presented. Also, exceptions or lack of correlations are pointed out. The authors show how their results agree or disagree with previously published papers, and discuss the theoretical implications as well as practical applications of the paper, and the significance of their results.

CONCLUSIONS

In Conclusions section the most important outcome of the work is stated, and interpretation of the findings also. If the authors have succeeded, or not, in addressing the need stated in the Introduction is reported here.

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If any product is cited in the manuscript the usage of ® or ™, and manufacturer data are mandatory. Use only scientific names of drugs. Include the manufacturer in brackets when describing equipment.

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Units of measurements should follow the primary language used (Portuguese/Spanish or English).

ABBREVIATIONS AND SYMBOLS

Abbreviations should follow the first mention of the term in the manuscript. The list of abbreviations is waived.

FOOTNOTES

Footnotes are used only in Tables/Boxes

03. USE OF COLORS

Although the use of color is permitted, it is important that authors (or professionals hired for editing) make an effort to ensure that the use of color does not impair understanding for readers with some form of visual impairment. We recommend consulting the following resources before preparing figures or tables using colors:

- How to make scientific figures accessible to readers with color-blindness (2019, Science News, The American Society for Cell Biology)
- Wong, B. Points of view: Color blindness. Nat Methods 8, 441 (2011).
<https://doi.org/10.1038/nmeth.1618>

04. FIGURES PREPARATION GUIDELINES

Graphs, photographs, diagrams, illustrations, and similar content should be referred to as figures (e.g., Figure 1, Figure 2, Figures 1, 2, 5-7) in ascending order according to their appearance in the text. Authors are strongly advised to adhere to the guidelines specified in the 'Use of Colors' and 'Preparation and Manipulation of Figures' sections below, in line with the International Committee of Medical Journal Editors (ICMJE) recommendations.

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ADDITIONALLY, WE STRONGLY ENCOURAGE AUTHORS (OR PROFESSIONALS HIRED FOR EDITING) TO OBSERVE AND FOLLOW THESE ADDITIONAL RECOMMENDATIONS:

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- Digital Images and Misconduct. (Council of Science Editors, White Paper on Publication Ethics)
- Preparing a Manuscript for Submission to a Medical Journal > Illustrations (Figures). (International Committee of Medical Journal Editors)

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Never build tables using spaces or tabs. Tables and Boxes must be created using the text editor built-in table creation tool and follow the following guidelines:

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- Avoid using colors to convey meaning, as screen readers and people with visual impairments may be disadvantaged. When absolutely necessary the use of colors should follow the guidelines in the "Use of colors" section.
- When using arrows, symbols (*, ‡, §, †, #, €, £, etc.), letters or numbers to include notes, be sure to clearly identify their use in the respective caption or table footnote.
- The use of decimal markers and the thousand separators must be observed and follow the text language.
- To maximize interoperability the use of diagonal cell splits is forbidden as these are not properly translated into XML and other electronic formats.

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Revision comments file	revisioncomments.pdf	Yes, when sending the revised version of your manuscript.

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The title page is a critical component of your submission and should include the following information, organized clearly and in order:

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Validation	Verification, whether as a part of the activity or separate, of the overall replication/ reproducibility of results/experiments and other research outputs
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S A V E T H E D A T E

INOVAÇÃO PARA A EVOLUÇÃO DA NEUROCIRURGIA



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CURITIBA | PR

Descriptive Study of the Brazilian Journal of Neurosurgery: a bibliometric analysis from 2002 to 2024

Estudo Descritivo do Brazilian Journal of Neurosurgery: uma análise bibliométrica de 2002 a 2024

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ABSTRACT

Introduction: The Brazilian Journal of Neurosurgery (JBNC), published by the Brazilian Academy of Neurosurgery, plays a vital role in advancing national science. **Objective:** This study aimed to conduct a bibliometric analysis of articles published in the JBNC to identify trends and characteristics in neurosurgery research. **Methods:** A bibliometric analysis was performed on articles published in the JBNC from 2002 to 2024. **Results:** A total of 866 articles were analyzed. Case reports were the most common type of publication, accounting for 42.3%, followed by original research at 28.5%. In terms of topics, brain tumors were the most frequently reported (23.9%), followed by vascular issues (22.9%) and spinal (12.1%). Male authors predominated, serving as first authors in 75.7% of cases and as last authors in 87.1% of cases. Additionally, the southeast region of Brazil contributed to 42.1% of all publications by Brazilian authors. **Conclusion:** The analysis revealed a regional and gender disparity in publications in the JBNC, with a concentration in the southeastern region and among male authors. These trends likely reflect the realities of the specialty, where approximately 50% of professionals are located in the southern region of Brazil, and 90% are male.

Keywords: Bibliometric analysis; Neurosurgery; JBNC

RESUMO

Introdução: O Jornal Brasileiro de Neurocirurgia (JBNC), publicado pela Academia Brasileira de Neurocirurgia, desempenha um papel vital no avanço da ciência nacional. **Objetivo:** Este estudo teve como objetivo realizar uma análise bibliométrica dos artigos publicados no JBNC para identificar tendências e características da pesquisa em neurocirurgia. **Métodos:** Foi realizada uma análise bibliométrica dos artigos publicados no JBNC entre 2002 e 2024. **Resultados:** Um total de 866 artigos foi analisado. Os relatos de caso foram o tipo mais comum de publicação, representando 42,3%, seguidos por pesquisas originais com 28,5%. Em relação aos temas, os tumores cerebrais foram os mais frequentemente abordados (23,9%), seguidos por doenças vasculares (22,9%) e coluna (12,1%). Autores do sexo masculino predominaram, sendo primeiros autores em 75,7% dos casos e últimos autores em 87,1%. Além disso, a região Sudeste do Brasil contribuiu com 42,1% de todas as publicações de autores brasileiros. **Conclusão:** A análise revelou uma disparidade regional e de gênero nas publicações do JBNC, com concentração na região Sudeste e entre autores do sexo masculino. Essas tendências provavelmente refletem a realidade da especialidade, na qual aproximadamente 50% dos profissionais estão localizados na região Sul do Brasil e 90% são homens.

Palavras-Chave: Análise bibliométrica; Neurocirurgia; JBNC

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INTRODUCTION

Each year, approximately 22.6 million people worldwide require neurological care due to comorbidities such as stroke, traumatic brain injury, and brain tumors, with over 60% of these cases requiring surgical intervention¹. As a result, neurosurgery has witnessed numerous advancements in diagnosis and treatment, largely driven by technological progress and supported by scientific research².

The Brazilian Journal of Neurosurgery (ISSN 0103-5118) was launched as the Seara Médica Neurocirúrgica in 1973. It is the official scientific journal of the Brazilian Academy of Neurosurgery (ABNc), published quarterly, and serves as an important communication platform³.

Bibliometric analysis is an analytical approach that evaluates scientific output using statistical and quantitative methods. It enables the identification of authors and research themes across various fields of knowledge, with origins dating back to the early 19th century⁴.

The objective of this study is to conduct a bibliometric analysis of the Brazilian Journal of Neurosurgery from 2002 to 2024.

METHODS

We conducted a bibliometric analysis of articles published in JBNC between 2002 and 2024. At least two different authors analyzed each article to minimize bias in data collection. Conference abstracts published as event summaries were excluded from the analysis.

The data analyzed from the articles included the following elements: 1) year of publication; 2) number of authors; 3) gender of the first and last authors; 4) country of the first and last authors (with states collected for authors from Brazil); 5) type of article (original research, review, case report, or brief note/other); 6) methodology (for original research, categorized as observational, randomized clinical trial, non-randomized clinical trial, guidelines/consensus, anatomical study, laboratory/experimental study, or experience report; for case reports, classified as single or series of cases—defined as

having two or more patients; for reviews, categorized as integrative, systematic, or systematic with meta-analysis); and 7) article topic (including vascular, spine, peripheral nerve, functional, pediatrics, brain tumors, trauma/neurointensive care, infectious/inflammatory, hydrodynamics, medical education, and other categories). An analysis between two periods was conducted to verify changes across the years (2002–2015 vs. 2016–2024). The division of years was carried out so that the proportion of articles was as close as possible between the periods.

The analysis was conducted using the Jamovi Project (2025), specifically Jamovi version 2.6 [Computer Software]. Data tabulation and analysis were performed with this software, while figures were created using GraphPad Prism 8.0 (GraphPad Software, San Diego, CA, USA). Continuous variables were summarized as means and standard deviations (SD), while categorical variables were presented as frequencies and percentages. For comparisons between the two periods, t-tests and chi-square tests were used, followed by post-hoc analysis with adjustment of residuals and verification of the alpha value, in order to identify statistically significant variables. In addition, simple linear regression was performed to assess the trend of quantitative variables over time. A p-value < 0.05 was considered statistically significant in all analyses, except in cases where post-hoc analysis correction was applied.

RESULTS

A total of 866 articles were published during the period (37.7 ± 16.4 papers/year), with an upward trend in the number of articles over the year (slope 2.1; $p < 0.001$) (Figure 1).

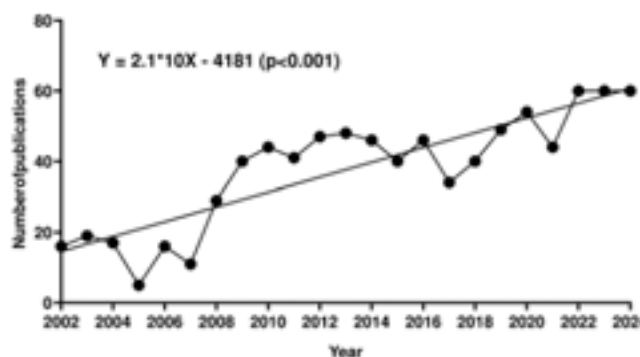


Figure 1. Number of publications in JBNC over time.

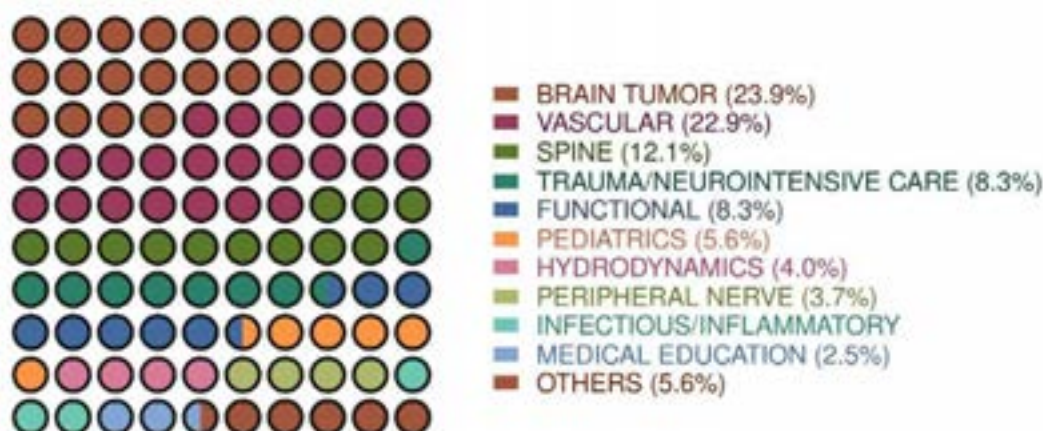


Figure 2. Percentage of publications categorized by the topic.

Case reports were the most common publications, accounting for 366 articles (42.3%), followed by original research articles (236, 27.3%), review articles (213, 24.6%), and brief notes/others (51, 5.9%). The main topics covered included brain tumors (195 articles, 23.9%), vascular diseases (187 articles, 22.9%), and spine disorders (99 articles, 12.1%) (Figure 2).

The data analyzed are presented in Table 1 (brief notes were excluded). On average, each publication had 5 ± 2 authors, with the numbers ranging from 1 to 18. Male authors were the most frequent, holding the positions as first and last author in 75.7% and 87.1% of the publications, respectively. In terms of methodology, observational studies were the most common among original articles (73.7%), while single case reports were the most frequent type among case reports (84.1%). Integrative reviews were the most frequent type among review articles (74.2%). A comparison of the different topics shows some relevant differences, such as the lower frequency of case reports and the higher proportion of females as primary authors in publications on functional neurosurgery, as well as a predominance of studies on trauma from the Northeast region (Table 1).

Analyzing the authors' affiliation (including brief notes), 90.1% of the publications were from Brazil, with most coming from the Southeast region (37.8%), followed by South (25.5%), Northeast (17.4%), Midwest (4.6%), and North (4.6%). São Paulo (230 papers; 29.5%), Paraná (130; 16.6%) and Rio Grande do Sul (62; 7.9%) were the states with most publications. The distribution by Brazilian states is shown in Figure 3. In terms of foreign affiliations, the United States (17 papers; 19.5% of foreign authors), Uruguay (17; 19.5%), and Portugal (6; 6.8%)

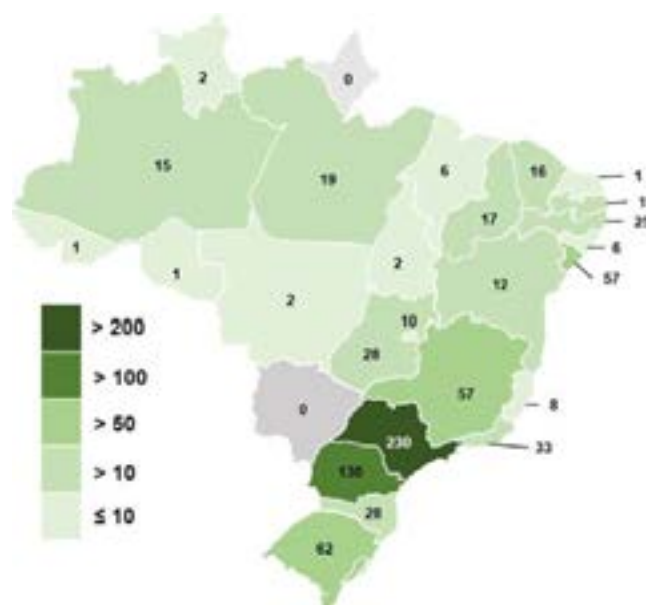


Figure 3. Geographical distribution in Brazil of the number of publications in the JBNC.

authored most publications. All continents were represented, except Oceania.

Comparing the two periods (2002–2015 vs. 2016–2024) presented in Table 2, it can be observed that, in the most recent interval, there was a higher average number of authors per article, a lower proportion of integrative reviews, an increase in systematic reviews, as well as greater female representation as first author—although less so as last author—and an increase in the participation of publications from the Midwest region.

Table 1. Data from articles published in JBNC divided by topic, except brief notes.

	Total	Brain tumor	Vascular	Spine	Functional	T. and n. care	Pediatrics	Hydrodynamics	P. nerve	Infect/inflam	M. education	Others
Number of publications	815	195 (23.9)	187 (22.9)	99 (12.1)	68 (8.3)	67 (8.3)	46 (5.6)	33 (4.0)	30 (3.7)	25 (3.1)	19 (2.5)	46 (5.6)
Number of authors	5 ± 2	5 ± 2	6 ± 2	5 ± 3	6 ± 3	5 ± 3	6 ± 3	5 ± 2	5 ± 2	6 ± 2	6 ± 4	5 ± 3
Type of study												
Review	213 (26.1)	45 (23.1)	44 (23.0)	26 (26.3)	33 (48.5)	16 (23.9)	9 (19.6)	10 (30.3)	5 (16.7)	-	8 (42.1)	18 (39.1)
Original	236 (29.0)	53 (27.2)	61 (33.1)	17 (17.2)	19 (27.9)	27 (40.3)	8 (17.4)	9 (27.3)	7 (23.3)	2 (8.0)	11 (57.9)	21 (45.7)
Case report	366 (44.9)	97 (49.7)	82 (43.9)	56 (56.5)	16 (23.6)	24 (35.8)	29 (63.0)	14 (42.4)	18 (60.0)	23 (92.0)	-	7 (15.2)
Methodology of the review												
Narrative	158 (74.2)	34 (75.5)	35 (79.5)	17 (65.3)	17 (51.5)	12 (75.0)	7 (78.0)	9 (90.0)	3 (60.0)	-	8 (100)	16 (88.8)
Systematic	48 (22.5)	10 (22.2)	8 (18.1)	8 (30.7)	13 (39.3)	3 (18.7)	1 (11.0)	1 (10.0)	2 (40.0)	-	-	2 (11.2)
Meta-analysis	8 (3.3)	1 (2.3)	1 (2.4)	1 (4.0)	3 (9.2)	1 (6.3)	1 (11.0)	-	-	-	-	-
Methodology of the original studies												
Observational	174 (73.7)	45 (84.9)	57 (93.4)	14 (82.3)	12 (63.1)	25 (93.0)	4 (50.0)	4 (44.4)	4 (57.1)	2 (100%)	4 (36.3)	3 (14.2)
Non-randomized clinical	9 (3.8)	2 (3.8)	1 (1.7)	1 (5.9)	2 (10.5)	-	1 (12.5)	1 (11.2)	1 (14.3)	-	-	-
Randomized clinical	3 (1.3)	-	-	-	1 (5.3)	1 (3.5)	1 (12.5)	-	-	-	-	-
Laboratory/experimental	8 (3.3)	-	-	1 (5.9)	2 (10.5)	-	1 (12.5)	2 (22.2)	1 (14.3)	-	-	1 (4.9)
Anatomical	26 (11.0)	-	2 (3.2)	1 (5.9)	1 (5.3)	-	1 (12.5)	2 (22.2)	-	-	4 (36.3)	15 (71.4)
Experience report	15 (6.9)	6 (11.3)	1 (1.7)	-	1 (5.3)	1 (3.5)	-	-	1 (14.3)	-	3 (27.3)	2 (9.5)
Type of case report												
Single	308 (84.1)	81 (83.5)	70 (85.3)	44 (78.5)	13 (81.2)	20 (83.3)	26 (89.6)	12 (85.7)	15 (83.3)	21 (91.3)	-	6 (85.7)
Series	58 (15.9)	16 (16.5)	12 (14.7)	12 (21.5)	3 (18.8)	4 (16.7)	3 (10.4)	2 (14.3)	3 (16.7)	2 (8.7)	-	1 (14.3)
Sex of the first author												
Male	617 (75.7)	153 (78.5)	137 (73.3)	85 (85.9)	41 (60.3)	50 (74.6)	30 (65.2)	28 (84.8)	23 (76.7)	23 (92.0)	14 (73.7)	33 (71.7)
Female	198 (24.3)	42 (21.5)	50 (26.7)	14 (14.1)	27 (39.7)	17 (25.4)	16 (34.8)	5 (15.2)	7 (23.3)	2 (8.0)	5 (26.3)	13 (28.3)
Sex of the last author												
Male	689 (87.1)	154 (83.2)	170 (92.4)	88 (90.7)	56 (86.2)	56 (83.6)	36 (78.3)	30 (93.8)	25 (89.3)	22 (88.0)	15 (83.3)	37 (84.1)
Female	102 (12.9)	31 (16.8)	14 (7.6)	9 (9.3)	9 (13.8)	11 (16.4)	10 (21.7)	2 (6.2)	3 (10.7)	3 (12.0)	3 (16.7)	7 (15.9)
First author from Brazil, divided by region	735 (90.1)	167 (85.6)	156 (83.4)	89 (89.9)	67 (98.5)	66 (98.5)	43 (93.5)	32 (97.0)	29 (96.7)	22 (88.0)	19 (100)	44 (95.7)
Southeast	302 (37.1)	66 (33.8)	64 (34.2)	45 (45.5)	36 (52.9)	19 (28.4)	10 (21.7)	20 (60.6)	12 (40.0)	11 (44.0)	4 (21.1)	15 (32.6)
South	211 (25.9)	62 (31.8)	53 (28.3)	17 (17.2)	12 (17.6)	14 (20.9)	13 (28.3)	5 (15.2)	6 (20.0)	5 (20.0)	8 (42.1)	16 (34.8)
Northeast	143 (17.5)	24 (12.3)	25 (13.4)	22 (22.2)	8 (11.8)	24 (35.8)	12 (26.1)	4 (12.1)	7 (23.4)	3 (12.0)	4 (21.1)	10 (21.7)
Midwest	39 (4.8)	6 (3.1)	7 (3.7)	1 (1.0)	10 (14.7)	4 (6.0)	5 (10.9)	2 (6.1)	1 (3.3)	-	2 (10.4)	1 (2.3)
North	39 (4.8)	9 (4.6)	7 (3.7)	4 (4.0)	1 (1.5)	5 (7.4)	3 (6.5)	1 (3.0)	3 (10.0)	3 (12.0)	1 (5.3)	2 (4.3)

Data are expressed as n (%) or as mean ± SD. Abbreviations: T. and n. care, Trauma/neurointensive care; P. nerve, Peripheral nerve; Intec/inflam, Infectious/inflammatory; M. education, Medical education.

Table 2. Data from articles published in JBNC divided by period, except brief notes.

	2002 - 2015	2016 - 2024	p value
Number of publications	411	404	
Number of authors	4.7 ± 2.2	6.1 ± 2.7	< 0.001
Type of study			
Review	112 (27.2)	102 (25.0)	NS
Original	127 (30.9)	108 (26.9)	
Case report	172 (41.9)	194 (48.1)	
Methodology of the review			
Narrative	104 (92.8)	54 (52.9)	< 0.001
Systematic	7 (6.2)	41 (36.6)	< 0.001
Meta-analysis	1 (1.0)	7 (10.5)	NS
Methodology of the original studies			
Observational	85 (66.9)	89 (82.4)	NS
Non-randomized clinical	7 (5.5)	2 (2.1)	
Randomized clinical	0	3 (2.7)	
Laboratory/experimental	5 (4.1)	3 (2.7)	
Anatomical	18 (14.1)	8 (7.4)	
Experience report	12 (9.4)	3 (2.7)	
Type of case report			
Single	140 (81.3)	168 (86.5)	NS
Series	32 (18.7)	26 (13.5)	
Sex of the first author			
Male	340 (82.7)	277 (68.5)	< 0.001
Female	71 (17.3)	127 (31.5)	
Sex of the last author			
Male	327 (84.0)	362 (90.0)	0.012
Female	62 (16.0)	40 (10.0)	
First author from Brazil, divided by region	359 (87.2)	375 (92.7)	NS
Southeast	167 (40.6)	135 (33.4)	NS
South	109 (26.5)	102 (25.2)	NS
Northeast	60 (14.6)	83 (20.5)	NS
Midwest	8 (1.9)	31 (7.7)	< 0.001
North	15 (3.6)	24 (5.9)	NS

Data are expressed as n (%) or as mean ± SD. Adjusted alpha for the first author from Brazil, divided by region = 0.004, and for the methodology of the original studies = 0.002. The numbers in bold represent values with statistical significance. Abbreviations: NS, not significant.

DISCUSSION

This bibliometric analysis of the Brazilian Journal of Neurosurgery (JBNC) reveals significant trends in the scientific production of neurosurgery in Brazil over 866 articles published in the past 23 years. The results show a marked increase in the number of publications over time, with case reports being more prevalent than original research and review articles. Brain tumors and vascular diseases emerged as the most frequently addressed topics. Additionally, the study highlights a notable gender disparity among first and last authors, as well as a regional concentration of publications in the South and Southeast regions of the country.

The increased number of publications in every year may reflect the journal's consolidation as a key platform for Brazilian neurosurgeons. This growth may be attributed to heightened awareness of the importance of research dissemination⁵, national incentives for academic productivity, and the expansion of graduate programs in neurosurgery and related fields⁶. Brazil currently has 3,643 neurosurgery specialists, compared to 1,735 in 2011, representing a 110% increase over this period⁷. This results in a rate of 1.71 specialists per 100,000 inhabitants, a number higher than the one estimated in most countries^{7,8}. Similarly, other specialties, such as rheumatology and transplantation medicine, have experienced significant growth in scientific output following the implementation of academic evaluation systems and greater literature visibility^{9,10}.

Case reports were the most common type of article, followed by original research and review articles. This publication profile is consistent with that of other specialty journals in developing countries, where limited research funding and infrastructure often result in a predominance of descriptive studies and case reports¹¹. While case reports are valuable for sharing rare or novel clinical experiences, the relatively low proportion of analytical and experimental studies may reflect structural barriers that hinder the production of higher-level evidence in the field¹². Nonetheless, the observed rise in systematic reviews in the second period (2016–2024) suggests a gradual improvement in methodological rigor and research training among contributors.

Authorship analysis revealed a consistent male predominance, with 75.7% of first authors and 87.1% of last authors being male, a pattern that remained stable across the two time periods analyzed.

Although this gender disparity reflects broader trends observed in surgical specialties, it warrants attention and mobilization^{13,14}. According to data from the 2025 Brazilian medical demography, 89.6% of Brazilian neurosurgeons are male, compared to 92.2% in 2015, indicating only modest progress in gender distribution over the past decade^{7,15}. Encouragingly, the second period showed a relative increase in female first authorship, suggesting a gradual improvement in gender equity within academic neurosurgery—an evolving trend also reported in the literature^{14,16}. The lower number of women as last authors may reflect their continued underrepresentation in senior academic and leadership roles within neurosurgery. Given that the last author typically holds the role of senior supervisor, this pattern suggests that, although female participation as first authors is increasing, many are still in the process of advancing to leadership roles, in parallel with the recent growth in the number of female neurosurgeons. Certain fields, such as functional neurosurgery, exhibited higher rates of female first authorship, possibly reflecting areas where female representation is gaining momentum. Institutional support and mentorship initiatives remain essential to further narrowing the gender gap in surgical research.

In terms of geographical distribution, the predominance of publications from the Southeast (37.8%) and South (25.5%) regions of Brazil is unsurprising, given the concentration of medical schools, research institutions, tertiary hospitals, and higher concentrations of neurosurgeons in these areas, a challenge observed worldwide as well^{17–19}. This imbalance highlights the persistent challenge of decentralizing academic neurosurgery in Brazil, a pattern also observed in different medical fields in the country^{9,20}. Currently, there is a significant concentration of neurosurgeons in specific areas, with 53.4% located in the Southeast region and also with 75.2% working in state capitals or cities with over 300,000 inhabitants⁷. However, the increased participation of authors from the Midwest region in the more recent period represents a positive sign of academic expansion and greater inclusivity, likely driven by the establishment of neurosurgery residency programs in previously underrepresented areas.

Thematic analysis identified brain tumors, vascular neurosurgery, and spine as the most frequent topics, aligned with global trends in neurosurgical research due to the high prevalence and clinical impact of these conditions^{21,22}. Interestingly, trauma-related articles showed a higher concentration in the Northeast, which may reflect local epidemiological demands and health system priorities²³. This regional specificity in research focus could be harnessed to develop targeted collaborative networks addressing public health needs in different areas of the country.

Despite the comprehensive scope of this study, some limitations must be acknowledged. First, the analysis relied exclusively on metadata and did not evaluate citation impact, scientific quality, or adherence to reporting guidelines, which limits the interpretation of the journal's academic influence. Second, the classification of article types and topics involved some degree of subjectivity, although it was mitigated by independent analysis by at least two reviewers. Finally, while the study spans more than two decades, the division into two periods for comparative analysis is arbitrary and may mask more nuanced temporal trends.

CONCLUSION

In summary, this bibliometric analysis highlights both significant progress and enduring disparities in Brazilian neurosurgical research. Despite a steady increase in publications—particularly after 2008, reflecting the impact of national research and graduate initiatives—marked gender and regional imbalances persist, with a predominance of male authorship and research concentrated in states with established academic centers such as São Paulo, Minas Gerais, and Rio de Janeiro. The high proportion of case reports compared with original research underscores structural limitations, including restricted funding and infrastructure, which hinder the production of higher-level evidence. Moving forward, strengthening academic neurosurgery in Brazil requires targeted actions to promote gender equity, geographic decentralization, thematic diversification, and methodological rigor. Investment in research training, international collaboration, and institutional incentives will be critical to fostering a more representative and clinically impactful scientific output.

REFERENCES

1. Dewan MC, Rattani A, Gupta S, et al. Estimating the global incidence of traumatic brain injury. *J Neurosurg.* 2018;130(4):1080-97. <http://doi.org/10.3171/2017.10.JNS17352>. PMID:29701556.
2. Muzumdar D. Neurosurgery in the past and future. An appraisal. *Ann Med Surg.* 2012;1:13-5. [http://doi.org/10.1016/S2049-0801\(12\)70004-3](http://doi.org/10.1016/S2049-0801(12)70004-3). PMID:26257899.
3. Brazilian journal of neurosurgery. About. Available from: <https://JBNC.org.br/about.php>. Accessed: 7/27/2025.
4. Donthu N, Kumar S, Mukherjee D, Pandey N, Lim WM. How to conduct a bibliometric analysis: an overview and guidelines. *J Bus Res.* 2021;133:285-96. <http://doi.org/10.1016/j.jbusres.2021.04.070>.
5. Carvalho ACC, Souza W. The evolution of Brazilian Health Sciences and the present situation. *Lancet Reg Health Am.* 2021;3:100044. <http://doi.org/10.1016/j.lana.2021.100044>. PMID:36777403.
6. Abdullah M, Ahmad B, Tufail Z, et al. Neurosurgery and social media: a bibliometric analysis of scientific production from 2004 to 2023. *World Neurosurg.* 2024;188:45-54. <http://doi.org/10.1016/j.wneu.2024.04.128>. PMID:38679373.
7. Scheffer M. Demografia médica no Brasil 2025. Brasília: Ministério da Saúde; 2025. 446 p.
8. Mukhopadhyay S, Punchak M, Rattani A, et al. The global neurosurgical workforce: a mixed-methods assessment of density and growth. *J Neurosurg.* 2019;130(4):1142-8. <http://doi.org/10.3171/2018.10.JNS171723>. PMID:30611133.
9. Cerqueira BP, Paim TS, Miyahara AK, et al. Estudo descritivo do Brazilian Journal of Transplantation: uma análise bibliométrica. *Braz J Transplant.* 2024;27(1):e2524. http://doi.org/10.53855/bjt.v27i1.579_PORT.
10. Xie X, Yu H, He Y, et al. Bibliometric analysis of global literature productivity in systemic lupus erythematosus from 2013 to 2022. *Clin Rheumatol.* 2024;43(1):175-87. <http://doi.org/10.1007/s10067-023-06728-z>. PMID:37668951.
11. Akhaddar A. Contribution of Moroccan neurosurgeons to the world neurosurgical data in PubMed: a bibliometric evaluation over time. *World Neurosurg.* 2019;126:59-65. <http://doi.org/10.1016/j.wneu.2019.02.122>. PMID:30844530.
12. Wallace SS, Barak G, Truong G, Parker MW. Hierarchy of evidence within the medical literature. *Hosp Pediatr.* 2022;12(8):745-50. <http://doi.org/10.1542/hpeds.2022-006690>. PMID:35909178.
13. Kim A, Dornelas LB, Telles L, et al. Brazilian authorship gender trends on academic surgery: a bigdata analysis. *Acta Cir Bras.* 2024;39:e397724. <http://doi.org/10.1590/acb397724>. PMID:39630701.
14. Aslan A, Kuzucu P, Karaaslan B, Börcek AÖ. Women in neurosurgery: gender differences in authorship in high-impact neurosurgery journals through the last two decades. *World Neurosurg.* 2020;138:374-80. <http://doi.org/10.1016/j.wneu.2020.03.017>. PMID:32200013.
15. Scheffer M, Cassenote A, Dal Poz MR, et al. Demografia médica no Brasil 2015. São Paulo: FMUSP; 2015. 284 p.
16. Smith BT, Villela-Castrejon J, Rodriguez L, et al. Authorship trends and gender concordance in surgical research. *J Surg Res.* 2025;312:148-54. <http://doi.org/10.1016/j.jss.2025.06.001>. PMID:40580574.
17. Koester SW, Bertani R, Batista S, et al. Current state of Brazilian neurosurgery: evaluation of burden of care and case volume. *World Neurosurg.* 2023;180:e309-16. <http://doi.org/10.1016/j.wneu.2023.09.066>. PMID:37769838.

18. Garg K, Chaurasia B, Gienapp AJ, Splavski B, Arnautovic KI. Bibliometric analysis of publications from 2011-2020 in 6 major neurosurgical journals (part 1): geographic, demographic, and article type trends. *World Neurosurg.* 2022;157:125-34. <http://doi.org/10.1016/j.wneu.2021.10.091>. PMID:34753011.
19. Santos RAD, Snell L, Nunes MDPT. Evaluation of the impact of collaborative work by teams from the National Medical Residency Committee and the Brazilian Society of Neurosurgery. Retrospective and prospective study. *Sao Paulo Med J.* 2015;134(2):103-9. <http://doi.org/10.1590/1516-3180.2015.9603001>. PMID:26465819.
20. Hoppen NHE, Vanz SAS. The development of Brazilian women's and gender studies: a bibliometric diagnosis. *Scientometrics.* 2023;128(1):227-61. <http://doi.org/10.1007/s11192-022-04545-w>. PMID:36467331.
21. Kanmounye US, Robertson FC, Sebopele LA, et al. Bibliometric analysis of the 200 most cited articles in world neurosurgery. *World Neurosurg.* 2021;149:226-231.e3. <http://doi.org/10.1016/j.wneu.2021.01.121>. PMID:33548539.
22. Ponce FA, Lozano AM. Highly cited works in neurosurgery. Part I: the 100 top-cited papers in neurosurgical journals. *J Neurosurg.* 2010;112(2):223-32. <http://doi.org/10.3171/2009.12.JNS091599>. PMID:20078192.
23. Gaudêncio TG, Leão GM. A epidemiologia do traumatismo crânio-encefálico: um levantamento bibliográfico no Brasil. *Rev Neurociências.* 2013;21(3):427-34. <http://doi.org/10.4181/RNC.2013.21.814.8p>.

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Ozonated Oil in Infected Surgical Wounds in Newborns with Dysraphisms: evaluation of adverse effects

Aceite Ozonizado En Heridas Quirúrgicas Infectadas en Recién Nacidos con Disrafismos: evaluación de efectos adversos

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ABSTRACT

Introduction: The Autonomous Institute of the University Hospital of the Andes (IAHULA) receives approximately 4 cases of spinal dysraphism and 0.3 cases of cranial dysraphism for surgical treatment each year, with a relatively high incidence of postoperative wound infection resulting in significant morbidity and mortality. **Objective:** The purpose of this study was to evaluate and monitor closure characteristics and possible adverse effects during the application of ozonated oil as adjuvant therapy in the treatment of infected surgical wounds in newborns with dysraphism. **Methods:** In this study, four newborns with dysraphisms were evaluated and followed: two spinal (myelomeningoceles) and two cranial (occipital encephaloceles) who underwent surgery. The surgical wounds were complicated by margin dehiscence associated with infection demonstrated by bacterial growth in exudate cultures. After obtaining the antibiogram report, each patient received parenteral treatment with specific antibiotics and a thin layer of ozonated oil on the wound surface. Body temperature, wound edge, adjacent skin characteristics, and complete blood count were monitored for three weeks. **Results:** As a result, infection was controlled in all wounds, hematological values normalized, no adverse skin adverse reactions were observed, and the wound healed satisfactorily. **Conclusion:** While a larger study is recommended, these results suggest that ozonated oil can be considered a relatively safe treatment for newborns with dysraphisms and infected surgical wounds.

Keywords: Dysraphisms; Surgical wound infection; Ozonated oil; Exudate cultures; Adverse reactions

RESUMEN

Introducción: El Instituto Autónomo del Hospital Universitario de los Andes (IAHULA) recibe aproximadamente 4 casos de disrafismo espinal y 0.3 casos de disrafismo craneal para tratamiento quirúrgico cada año con una incidencia relativamente alta de infección de herida postoperatoria resultando en morbilidad y mortalidad significativas. **Objetivo:** El propósito de este estudio fue evaluar y monitorear las características de cierre y los posibles efectos adversos durante la aplicación de aceite ozonizado como terapia adyuvante en el tratamiento de heridas quirúrgicas infectadas en recién nacidos con disrafismo. **Métodos:** En este estudio, cuatro recién nacidos con disrafismos fueron evaluados y seguidos: dos espinales (mielomeningoceles) y dos craneales (encefaloceles occipitales) sometidos a cirugía. Las heridas quirúrgicas se complicaron por dehiscencia de márgenes asociada a infección demostrada por crecimiento bacteriano en cultivos de exudado. Luego de obtener el reporte del antibiograma, cada paciente recibió tratamiento parenteral con antibióticos específicos y una capa delgada de aceite ozonizado sobre la superficie de la herida. Se monitorizaron la temperatura corporal, el borde de la herida, las características de la piel adyacente y un hemograma completo durante tres semanas. **Resultados:** Como resultado, se controló la infección en todas las heridas, los valores hematológicos se normalizaron, no se observaron reacciones cutáneas adversas y la herida cicatrizó satisfactoriamente. **Conclusión:** Si bien se recomienda un estudio más amplio, estos resultados sugieren que el aceite ozonizado puede considerarse un tratamiento relativamente seguro para recién nacidos con disrafismos y heridas quirúrgicas infectadas.

Palabras-Clave: Disrafismos; Infección de herida quirúrgica; Aceite ozonizado; Cultivos de exudado; Reacciones adversas

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INTRODUCTION

Both encephalocele and myelomeningocele represent developmental disorders classified as neural tube defects of the dysraphism type¹. Although values can vary significantly depending on the region, It has been reported that incidence of congenital encephalocele estimated at 1 in 10,000 live births², and Myelomeningocele incidence is approximately 0.6 cases per 1000 live births³.

Newborns with dysraphism are admitted to the Neonatal Care Unit of the Autonomous Institute of the University Hospital of the Andes (IAHULA) and are evaluated by the Neurosurgery Unit. According to our records, approximately 4 cases of spinal dysraphism and 0.3 cases of cranial dysraphism are surgically treated each year. The incidence of postoperative wound infections is relatively high, resulting in significant morbidity and mortality. The main pathogens isolated in the IAHULA neonatal unit include *Klesiella sp.*, *Klesiella pneumoniae*, *Klesiella aerogenes*, *Pseudomona aeruginosa*, *Escherichia coli*, and *Staphylococcus aureus*. These germs are involved in complications such as: suture dehiscence, fever, and the spread of infection to the central nervous system, among others.

The partial or total separation of the previously approximated wound edges when healing in its early stages is known as dehiscence and among the causes are infection⁴. A wound infection prolongs hospitalization time, increases the consumption of medications and antibiotics, and delays healing⁵.

In our hospital, the characteristics of an infected wound after surgery must be recognized early to avoid complications, especially in newborns with an immature immune system. Furthermore, the risk is greater in patients who present open dysraphism.

It has been reported that the oxidation of fatty acids present in oils (such as: olive, sesame, peanut, coconut, theobroma, soybean and jojoba oils) during the ozonation reaction has the capacity to produce lipoperoxides and ozonides and the products of these reactions seem to have germicidal and immunostimulant properties as well as favor tissue repair⁶. In a work published by Godínez and collaborators, ozonized vegetable oils were shown to have a high antibacterial capacity against *L. monocytogenes*⁷.

In relation to the adverse effects reported with the use of ozonide oils, the following stand out very low, 0.3% mainly: maceration, desquamation, fissures, erythema, vesicles and itching, burning sensation, and edema⁸.

On the basis of the proposed germicidal, immune-stimulating, and tissue-regenerating action, topical application to the wound is proposed with prior authorization following the hospital's ethics committee protocols. Informed consent was obtained from the newborns' parents and guardians for the therapeutic application of ozonide oil.

The purpose of this study was to evaluate and monitor closure characteristics and possible adverse effects during the application of ozonized oil as adjuvant therapy in the treatment of infected surgical wounds in newborns with dysraphism.

METHODS

During postoperative follow-up, samples are taken for bacteriological culture in newborns with suspected surgical wound infection.

Four patients with dysraphism were selected (Patient 1 – Figure 1, Patient 2 – Figure 2, Patient 3 – Figure 3, and Patient 4 – Figure 4): two cranial (Figures 1A and 2A) and two spinal (Figures 3A and 4A). Five days after surgery, an infectious complication of the surgical wound was detected. Informed verbal and written consent were obtained from the newborn's parents and guardians (according to the hospital ethics committee) to receive adjuvant topical therapy. All patients initially received postoperative pharmacological therapy with intravenous antibiotics — prophylactic antibiotics — and then were adjusted according to the antibiogram.

Each patient received a daily bath and a combination of formula and breastfeeding. Following the bath, these patients received daily wounds and adjacent skin care exclusively with saline solution (0.9% NaCl saline). Once a day, for a total of three weeks, after washing, the wound was gently dried and a thin layer of ozonized oil was applied to the entire surface (volume 1 cm³ approximately



Figure 1. Patient 1. A-C. Cranial dysraphism (occipital encephalocele) evolution. A. Presurgical. B. Postoperative wound infection with *Klesiella pneumoniae*. C. Wound in the third week.



Figure 2. Patient 2. A-C. Cranial dysraphism (occipital encephalocele) evolution. A. Presurgical. B. Postoperative wound infection with *Staphylococcus aureus*. C. Wound in the third week.



Figure 3. Patient 3. A-C. Spinal dysraphism (myelomeningoceles) evolution. B. Postoperative wound infection with *Pseudomona aeruginosa*. C. Wound in the third week.

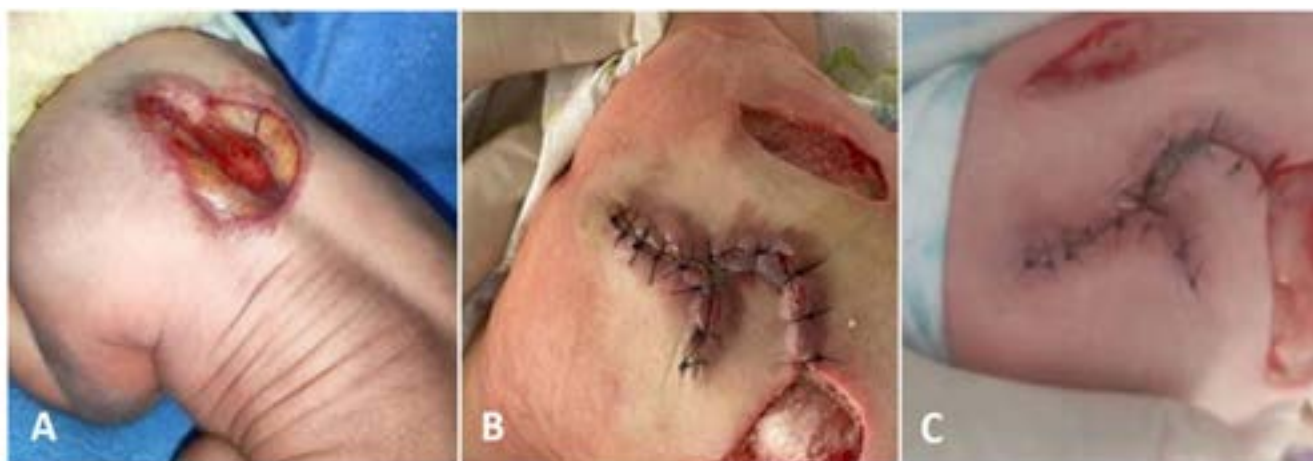


Figure 4. Patient 4. **A-C.** Spinal dysraphism (myelomeningoceles). **A.** Presurgical. **B.** Postoperative wound infection with *Streptococcus pneumoniae*. **C.** Wound in the third week.

a concentration of 4.24 mg/gr) on sterile gauze. The ozonized oil used was a mixture of vegetable oil and ozone (O₃). The main chemical component is vegetable oil, along with stable oxygenated compounds, which react with the oil, to determine the formation of ozonides and peroxides.

Each week, the presence of healing bridges between the wound edges was evaluated. An examination was performed to verify changes in skin color, the presence of areas of suppuration, or local volume increases. Fever was monitored, and a complete blood count was performed. Images were taken.

RESULTS

The bacterial microorganisms isolated from infected postoperative wounds included *Klesiella pneumoniae* (Figure 1B), *Staphylococcus aureus* (Figure 2B), *Pseudomonas aeruginosa* (Figure 3B), and *Streptococcus pneumoniae* (Figure 4B). Patients received parenteral antibiotic therapy at a meningeal dose. Furthermore, the closure of stable wounds treated with topical ozonized oil was observed in all the patients. Local control of the infection was observed. No patients presented with fever or meningeal syndrome. The bridges between surgical wound edges were consolidated by the end of the third week of treatment. Routine laboratory tests, such as acute phase reactant and complete blood count, revealed progression to a stable level considered normal.

Skin sutures were made with nylon monofilaments and their removal began after the third week of treatment. All patients were discharged in good clinical condition and underwent consolidated wound closure (Figures 1C, 2C, 3C, and 4C).

DISCUSSION

Newborns with encephalocele and myelomeningocele are presented, undergoing surgery and postoperative wound follow-up. The incidence of surgical site infection is high, including the separation of previously approximated edges, as indicated by Rosen and Manna⁴. Manifestations of infection such as dehiscence of the surgical wound suture, skin redness, local warmth, fever, and neutrophilia in the complete blood count were associated with the isolation of a specific bacterium in culture of wound edge secretion.

Although Godínez et al. demonstrated that ozonized vegetable oils have a high antibacterial capacity against *L. monocytogenes*⁷, the present study confirmed their effective efficacy against multiple microorganisms, i.e., the product offers a broad bacterial spectrum.

In general, complications of this infectious process affecting the nervous system are expected, but in our cases, seizures or meningismus did not occur. In three patients, a ventriculoperitoneal shunt was placed due to previous diagnoses of hydrocephalus. During topical application of ozonized oil, no adverse skin

reactions were observed, and according to the literature reviewed, this topical application could improve wound characteristics.

CONCLUSION

This study suggests that the topical use of ozonized oil can be considered an adjuvant option for the treatment of complicated surgical wounds in newborns with dysraphism and appears to be a safe product. However, additional studies with larger sample sizes are needed to validate the therapeutic indications.

REFERENCES

1. Salih MA, Murshid WR, Seidahmed MZ. Classification, clinical features, and genetics of neural tube defects. *Saudi Med J*. 2014;35(Suppl 1):S5-14. PMID:25551113.
2. Matos Cruz AJ, De Jesus O. Encephalocele. *Treasure Island: StatPearls Publishing*; 2024. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK562168/>. Accessed: 5/7/2025.
3. El Halaby WE, Ismail MT. Delayed hydrocephalus after repairing un-rupture myelomeningocele. *Egypt J Neurosurg*. 2016;31(3):167-70.
4. Rosen RD, Manna B. Wound dehiscence. *Treasure Island: StatPearls Publishing*; 2023. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK551712/>. Accessed: 5/7/2025.
5. Hajmohammadi K, Mohammadpour Y, Parizad N. Fighting postsurgical infection after myelomeningocele repair with medical honey (Medihoney): a case report. *Childs Nerv Syst*. 2023;39(7):1969-76. <http://doi.org/10.1007/s00381-023-05929-w>. PMID:36959360.
6. Sánchez GM, Re L, Davison GP, Delaporte RH. Las aplicaciones médicas de los aceites ozonizados: actualización. *REO*. [Internet]. 2012 [Accessed: 5/7/2025];2(1):121-39. Available from: <https://www.researchgate.net/publication/277042731>
7. Godínez-Oviedo A, Zamora-Rodríguez Z, Martínez-Juárez V, Fleitas-González E, Hernández-Rosado A, Peña-Jiménez F. Evaluación

del efecto antibacterial del aceite de oliva ozonizado contra *Listeria monocytogenes*. *Abanico Veterinario*. 2017;7(1):36-43. <http://doi.org/10.21929/abavet2017.71.3>.

8. Menéndez S, Re L, Falcón L, et al. Safety of topical Oleozon® in the treatment of tinea pedis: phase IV clinical trial. *Int J Ozone Therapy*. 2008;7(1):55-9.

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Use of 3D Printing Technology in the Localization of Intracranial Lesions and Craniotomy Planning

Utilização da Tecnologia de Impressão 3D na Localização de Lesões Intracranianas e Planejamento de Craniotomia

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ABSTRACT

Introduction: accurate localization of intracranial lesions is essential in modern neurosurgery. **Objective:** this study proposes the validation of a device produced using 3D printing to assist in identifying the central point of deep brain lesions and in planning the craniotomy. **Methods:** imaging studies were used for segmentation, modeling software, and 3D printing, and accuracy was compared to intraoperative neuronavigation. **Results:** the device demonstrated acceptable error and clinical feasibility, especially in centers with limited resources. **Conclusion:** This technology demonstrated an effective alternative for accurately determining the center point of a lesion and delimiting the craniotomy area, regardless of the availability of neuronavigation or stereotaxis.

Keywords: 3D printing; Lesion localization; Craniotomy; Neuronavigation; Surgical planning; Neurosurgery

RESUMO

Introdução: a localização precisa das lesões intracranianas é essencial na neurocirurgia moderna. **Objetivo:** este estudo se propõe a validar um dispositivo produzido com impressão 3D para auxiliar na identificação do ponto central de lesões cerebrais profundas e no planejamento da craniotomia. **Métodos:** estudos de imagem foram utilizados para segmentação e modelagem para impressão 3D, e a precisão foi comparada à neuronavegação intraoperatória. **Resultados:** o dispositivo mostrou erro aceitável e viabilidade clínica, especialmente em centros com recursos limitados. **Conclusão:** esta tecnologia demonstrou ser uma alternativa eficaz para determinar com precisão o ponto central de uma lesão e delimitar a área de craniotomia, independentemente da disponibilidade da neuronavegação ou estereotaxia.

Palavras-Chave: Impressão 3D; Localização da lesão; Craniotomia; Neuronavegação; Planejamento cirúrgico; Neurocirurgia

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INTRODUCTION

The localization of cortical and subcortical lesions in neurosurgery without adequate planning carries a high margin for error, often resulting in the need for an expanded craniotomy and, consequently, greater surgical morbidity. In modern neurosurgery, such outcomes are considered unacceptable given the extensive knowledge and technological resources available^{1,2}.

Techniques used to aid in the correct localization of cortical and subcortical lesions include craniometric points, stereotaxy, and neuronavigation, each with its own advantages and limitations. Stereotaxy and neuronavigation are well-established technologies supported by the literature, but due to their availability and cost, they are not widely accessible to most neurosurgeons³.

In this context, 3D printing (3DP) emerges as a tool of great potential, enabling the creation of personalized anatomical models—not only for teaching purposes but also directly applicable to surgical planning—that can be used for the production of prostheses, implants, and surgical guides^{4,5}. Previous work by our group has shown that the use of such biomodels contributes to improved anatomical understanding, individualized planning, reduced operative time, and potential reduction in surgical morbidity⁶⁻¹⁰.

However, the literature still lacks specific studies on the use of medical image segmentation and additive manufacturing for lesion localization in neurosurgery. The present study aims to present a low-cost device that determines the central point of an intracranial lesion — particularly cortical and subcortical lesions — while delimiting the craniotomy area^{3,7}. Additionally, by relying on low-cost technology already validated in other settings by our team, this proposal reinforces the feasibility of 3DP as an accessible and applicable tool in multiple neurosurgical contexts³.

METHODS

Image acquisition

Images were obtained via 1.5 Tesla Magnetic Resonance Imaging (MRI) and computed tomography (CT) at a neurosurgery hospital in Curitiba, Brazil, approximately one month before the procedure.

Image registration and segmentation

Multimodal registration of brain MRI (cube FLAIR sequence) and CT scans were performed using the 3D Slicer software¹¹ (version 5.8.1). The General Registration (BRAINS) module with rigid transformation (6 degrees of freedom) was used (Figure 1).

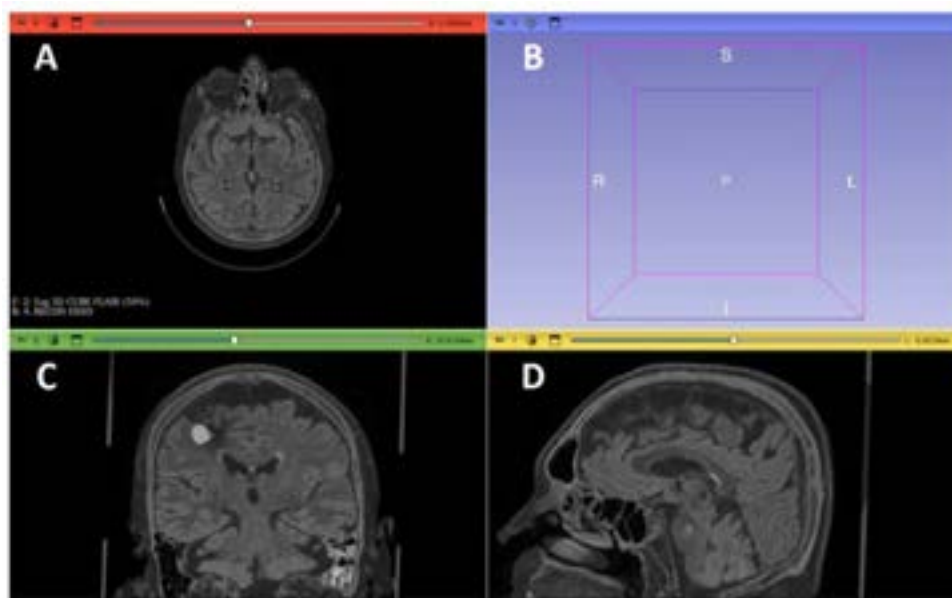


Figure 1. A, B, C and D. Three-plane view of the MRI-CT overlay after registration.



Figure 2. A, B, C and D. View showing segmentation masks: tumor (red), skull (blue), and external facial contour (yellow).

Subsequently, semi-automatic segmentation of the structures of interest was performed using the Segment Editor module. The tumor was delineated using the region growing technique with individually adjusted intensity thresholds, while the skull and external facial contour were segmented using an intensity range-based approach. Segmentations were manually refined by an experienced neurosurgeon to ensure anatomical accuracy (Figure 2).

Locator modeling

After segmentation of the anatomical structures (tumor, skull, and craniofacial contour), digital models were exported from 3D Slicer® in STL format. The surgical locator was modeled in Autodesk Meshmixer® software (version 3.5, Autodesk Inc., San Rafael, CA). STL files were imported into Meshmixer®, and digital artifacts were removed using the Inspector tool (tolerance: 0.1 mm).

Support points for the locator were defined at the tragus (ear) and glabella (nose). Modeling was performed using selection, extrusion, and sculpting tools. The locator's central point was defined by generating a cylinder with an angle adjusted along the access axis. Figure 3 illustrates the device modeling.

3D Printing of the Locator

The device was manufactured using a Bambu Lab X1 Carbon 3D printer (Bambu Lab, China) with fused filament fabrication (FFF)

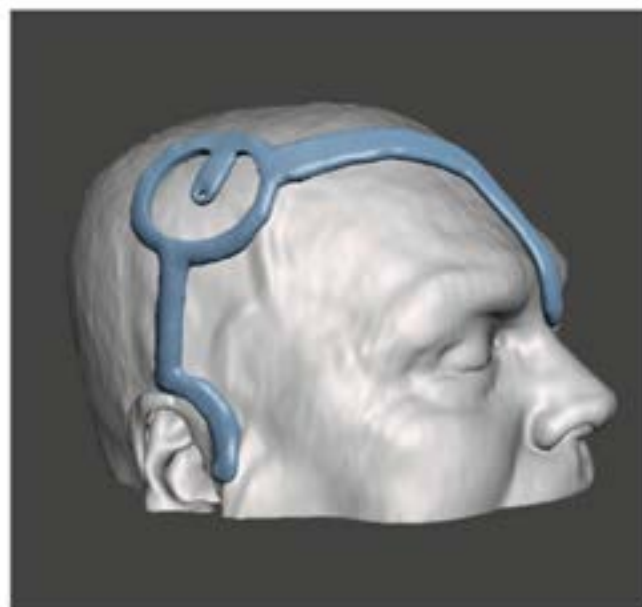


Figure 3. Modeling of the custom surgical locator in Autodesk Meshmixer®.

technology. The filament used was PLA (polylactic acid), 1.75 mm in diameter, with printing parameters: extrusion temperature 220°C, bed temperature 60°C, layer height 0.2 mm, and printing speed 200 mm/s. Post-processing consisted of support removal, surface sanding, and visual inspection to ensure the absence of critical imperfections.

RESULTS

After placement on the patient's head, it was verified through Medtronic® neuronavigation that the central point indicated by the locator matched the neuronavigation-defined lesion point. Thus, the device proved to be a low-cost and highly user-friendly tool.

Case 1

Patient, 76 years old, diagnosed with a cavernoma in the right middle frontal gyrus. The device was designed to be fitted along the midline and a lateral line, using the patient's ear as a reference. The device's circular outline represented the craniotomy area, with the central point of the lesion located within it.

Neuronavigation confirmed the match between the locator's central point and the lesion's central point, enabling the procedure to proceed safely without the risk of marking errors. After craniotomy and cavernoma identification, the device's accuracy was confirmed to be comparable to neuronavigation in lesion localization and craniotomy planning (Figures 4 and 5).

Case 2

Patient, 27 years old, presented with seizures and was diagnosed with a left frontal cavernoma. Lesion resection was indicated, and a new 3D-printed locator device was used to assist in cavernoma localization (Figures 6 and 7). Correspondence with neuronavigation was verified, enabling safe craniotomy planning.

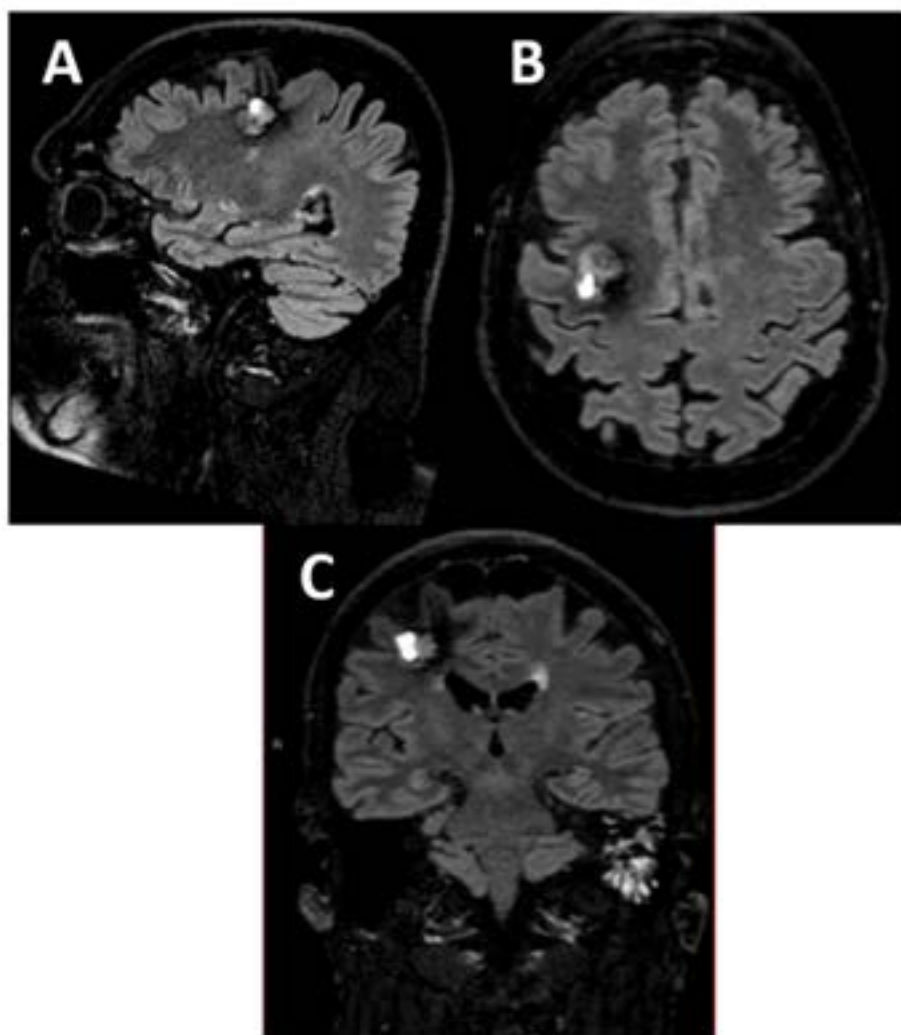


Figure 4. A, B and C. FLAIR-weighted Magnetic Resonance Imaging showing a cavernoma in the right superior frontal gyrus.



Figure 5. A and B. Photograph demonstrating the locator model on the patient, where there is a central point representing the correspondence with the central point of the lesion on the surface and a circumference around the central point representing the craniotomy area to be performed.

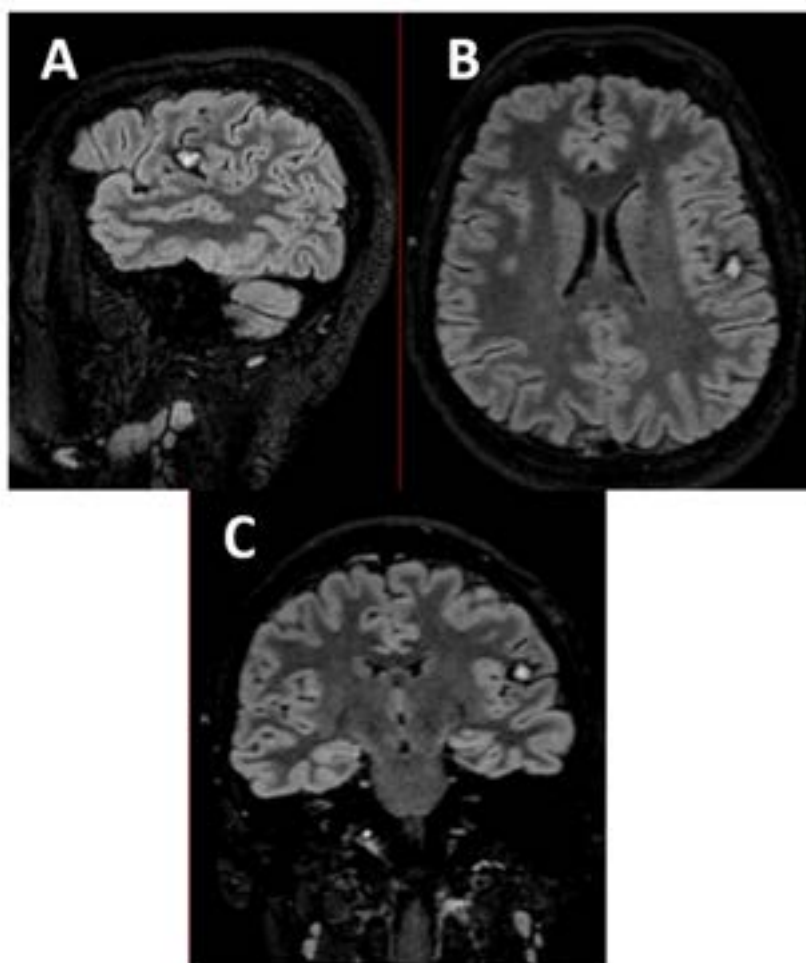


Figure 6. A, B and C. FLAIR-weighted Magnetic Resonance Imaging showing a cavernoma in the left inferior frontal gyrus.

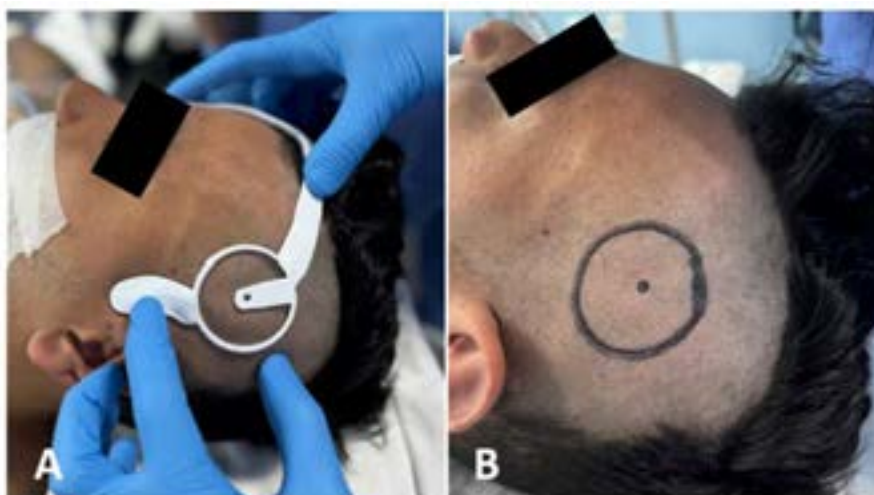


Figure 7. A and B. Photograph demonstrates the locator model on the patient, which also represents the central point of the lesion on the surface and a circumference around the central point representing the craniotomy area to be performed.

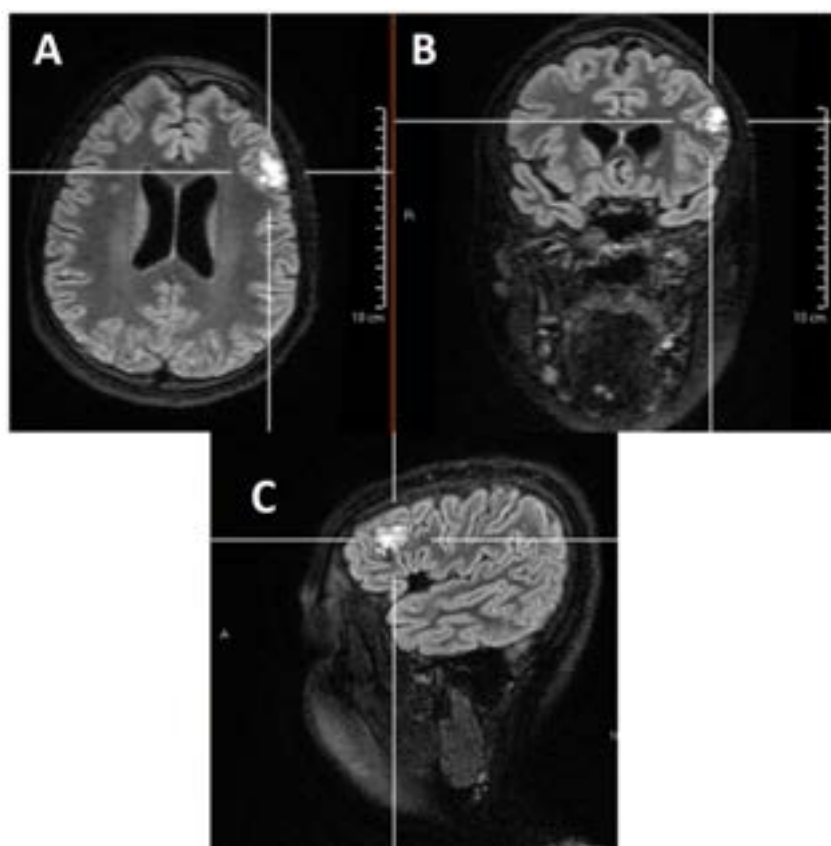


Figure 8. A, B and C. FLAIR-weighted Magnetic Resonance Imaging showing a cavernoma in the left middle frontal gyrus.

Case 3

Patient, 18 years old, presented with recurrent seizures and was diagnosed with a left frontal cavernoma. Resection of the lesion was indicated, using a localizing device with reference points

at the midline and a lateral line based on the ear. Once again, there was adequate correspondence with neuronavigation, allowing for safe planning and performance of the craniotomy (Figures 8 A,B and 9 A,B).

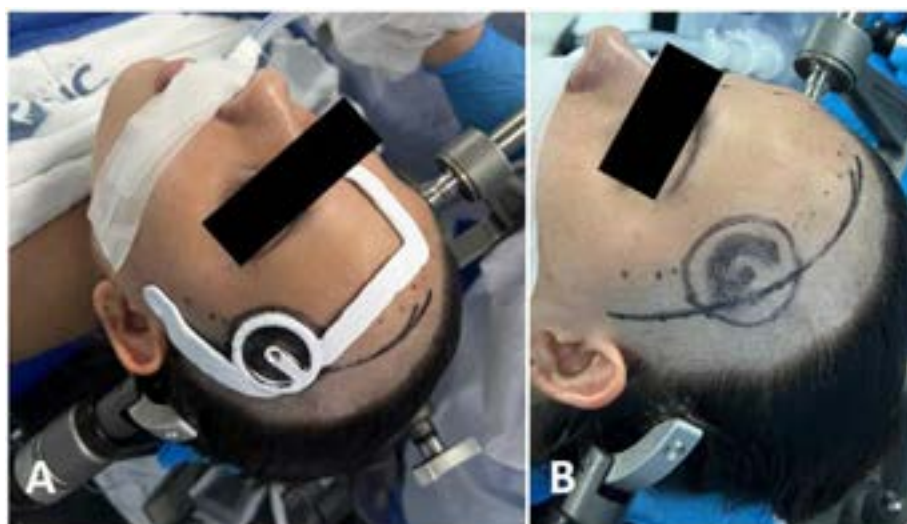


Figure 9. A and B. Photograph demonstrates the locator model on the patient. The central point of the lesion on the surface and a circumference around the central point representing the craniotomy area to be performed.

DISCUSSION

Neurosurgical planning for cortical and subcortical lesions remains a significant challenge given the brain's anatomical complexity and the need for precision to minimize patient risk. Classical craniometric points often have limitations: they may present individual variations, be poorly visible during surgery, or prove insufficient for access to complex regions. Furthermore, while neuronavigation is desirable, its availability remains restricted in many centers, especially in resource-limited regions^{3,9}.

In this scenario, it is essential to seek accessible and effective solutions capable of replacing high-cost technologies without compromising surgical safety or outcomes. While there are increasing reports on the use of biomodels for simulation and training—particularly for vascular pathologies and tumors—there are still few publications focusing on their intraoperative use for lesion localization, highlighting the relevance of the present investigation^{1,4}.

The results we obtained from Cases 1 and 2 demonstrate the ability of the locator device to accurately determine the central point of intracranial lesions and delimit the craniotomy area, playing a role as important as a neuronavigator in these aspects. Both cases involved lesions that could have significant

localization errors if guided solely by anatomy. Thus, the low-cost and easy-to-use device represents a simple way to democratize precision neurosurgery⁷.

Custom surgical guides produced via 3D printing are already used in neurosurgery and neuroendoscopy^{10,12,14}. For example, Yavas et al. (2021) demonstrated the use of 3DP in creating augmented reality markers adapted to neuronavigation. In a recent study, Zhou et al¹⁴ employed 3DP to produce a device specifically for locating parasagittal meningiomas, proving to be a valuable, low-cost solution with potential application for other intracranial tumors⁹.

In this context, our study demonstrated the feasibility of using an equally accessible and easy-to-handle 3D-printed locator device, representing a highly relevant auxiliary tool for craniotomy planning—particularly in environments where neuronavigation and stereotaxy are scarce or unavailable. Main advantages include ease of use, reduced need for specialized training, minimal learning curve, and immediate incorporation into neurosurgical routines⁷.

Even with promising results, there are still some limitations in our study that should be considered. We must take into account a limited number of cases, which restricts the generalization of findings, requiring validation in studies with larger and more diverse samples. Furthermore, the dependence on image quality and the

accuracy of manual segmentation is also a critical factor, requiring rigorous protocols and validation by experienced neurosurgeons¹⁰.

Adopting this resource can significantly expand access to precision neurosurgery in resource-constrained environments, without burdening healthcare services or generating additional costs for patients, contributing to greater equity and quality in surgical outcomes⁹. As future perspectives from this study, we should focus on validating the locator with larger numbers and diversification of patients, covering different types of intracranial lesions.

CONCLUSION

Our study proposes the use of a low-cost 3D-printed locator device suitable for both high- and low-complexity environments³. The device proved to be an effective alternative for accurately determining the central point of a lesion and delimiting the craniotomy area, regardless of neuronavigation or stereotaxy availability.

REFERENCES

1. Cogswell PM, Rischall MA, Alexander AE, Dickens HJ, Lanzino G, Morris JM. Intracranial vasculature 3D printing: review of techniques and manufacturing processes to inform clinical practice. *3D Print Med*. 2020;6(1):18. <http://doi.org/10.1186/s41205-020-00071-8>. PMID:32761490.
2. Sun Z, Liu D, Samoudi A, Nguyen D, Houssein S. Three-dimensional printing in neurosurgery: a review of current applications. *J Clin Neurosci*. 2018;50:1-7. <http://doi.org/10.1016/j.jocn.2018.01.050>.
3. Nizar A, Gerganov VM, Samii M. Low-cost and open-source three-dimensional (3D) printing in neurosurgery: A pilot experiment using direct drive modification to produce multi-material neuroanatomical models. *J Neurosurg*. 2023;140(4):1091-101. <https://doi.org/10.1016/j.clineuro.2023.107684>. PMID:37890179.
4. Martelli N, Serrano C, van den Brink H, et al. Efficacy of patient-specific 3D-printed tumor models in surgical planning. *J Neurosurg*. 2019;130(2):491-5.
5. Dering LM, Fernandes BL, Pedro MKF, Leal AG, Souza MA. 3D and 4D printing for biomedical applications. In: Gupta RK, editor. *3D printing*. 1st ed. Boca Raton: CRC Press; 2023. p. 325-38. <http://doi.org/10.1201/9781003296676-21>.
6. Dering LM, Pedro MKF, Silva ACF, Leal AG, Souza MA. Experience in using additive manufacturing of cerebral aneurysms as a 3D assistant tool in surgical planning. *Braz Arch Biol Technol*. 2022;65:e22210575. <http://doi.org/10.1590/1678-4324-2022210575>.
7. Leal AG, Dering LM, Pedro MKF, Fernandes BL, Souza MA, Nohama P. Aplicações clínicas de modelos de manufatura aditiva em neurocirurgia. In: Leal AG, Aguiar PHP, Ramina R, editors. *Tratado de neurologia clínica e cirúrgica*. 1st ed. Ponta Grossa: Atena; 2022. Vol. 1, p. 853-68. <http://doi.org/10.22533/at.ed.34622130462>.
8. Silva ACF, Dering LM, Pedro MKF, Montibeller GR, Leal AG. Planejamento pré-operatório por meio de manufatura aditiva em crânioestenose. *J Bras Neurocir*. 2022;33(1):127-32. <http://doi.org/10.22290/jbnc.2022.330102>.
9. El Hussein H, Silva JFC, Leal AG, et al. Dural closure training with prototyped model. *Cureus*. 2024;16(6):e61688. PMID:38975561.
10. Hungria EB, Dering LM, Lima MER, Pedro MKF, Leal AG. Development of prototyped molds for cranioplasty using desktop 3D printers: a single-center experience. *World Neurosurg X*. 2025;27:100479. <http://doi.org/10.1016/j.wnsx.2025.100479>.
11. 3D SLICER SOFTWARE. 3D Slicer image computing platform. Available from: www.slicer.org. Accessed: 7/27/2025.
12. Yavas G, Caliskan KE, Cagli MS. Three-dimensional-printed marker-based augmented reality neuronavigation: a new neuronavigation technique. *Neurosurg Focus*.
13. Yavas G, Caliskan KE, Cagli MS. Three-dimensional-printed marker-based augmented reality neuronavigation: a new neuronavigation technique. *Neurosurg Focus*. 2021 Aug;51(2):E20. doi: 10.3171/2021.5.FOCUS21206.
14. Zhou L, Wang W, Li Z, Wei H, Cai Q, Chen Q, et al. Clinical application of 3D-Slicer + 3D printing guide combined with transcranial neuroendoscopic in minimally invasive neurosurgery. *Sci Rep*. 2022 Nov 28;12(1):20421. doi: 10.1038/s41598-022-24876-1.

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Clinical Epidemiological Profile of Patients with Cerebral Metastasis

Perfil Clínico Epidemiológico de Pacientes com Metástase Cerebral

Débora de Melo Cardoso¹ 

Carlos Umberto Pereira^{2,3} 

Samuel Pedro Pereira Silveira⁴

ABSTRACT

Introduction: Cerebral metastasis (CM) is the major neurological manifestation of cancer. Most prevalent primary sites include: lungs, breast and skin. Hemiparesis, aphasia, visual disturbances, seizures, headache, nausea, vomiting and cognitive abnormalities are part of the clinical manifestations. Imaging studies associated with biopsy confirms the diagnosis. The treatment may be palliative or surgical and complemented by radiotherapy and chemotherapy. **Objective:** To describe the clinical and epidemiological characteristics of CM in cancer patients. **Methods:** A retrospective, descriptive and cross-sectional study of confirmed cases of CM admitted to the neurosurgery service of the Emergency Hospital of Sergipe (Aracaju-Se) was conducted, from January 2011 to August 2012. **Results:** Forty patients with a mean age of 53.2 years were studied. The sixth decade of life was the most affected age group. Regarding gender, 24 (60%) were female and 16 (40%) male. The most prevalent primary site of CM was breast cancer (37.5%), followed by lung, 30%. The most common symptoms were headache and cognitive abnormalities both present in 30 (75%) cases. A computed tomography (CT) was the most used diagnostic method, 36 (90%) patients. Palliative treatment was carried out in 25 (62.5%) patients, followed by radiotherapy, 11 (27.5%), surgery, 8 (20%) and chemotherapy, 3 (7.5%) patients. **Conclusion:** The clinical and epidemiological analyses of cancer patients is extremely important for the diagnosis of CM.

Keywords: Neoplasm metastasis; Brain neoplasm; Epidemiology; Neurologic manifestations

RESUMO

Introdução: A metástase cerebral (MC) é a principal manifestação neurológica do câncer. Sítios primários mais prevalentes incluem: pulmão, mama e pele. Hemiparesia, afasia, distúrbio visual, crises convulsivas, cefaleia, náusea, vômito e anormalidade cognitiva fazem parte do quadro clínico. Exames de imagem associado à biópsia confirmam o diagnóstico. O tratamento pode ser paliativo ou cirúrgico e complementar com radioterapia e quimioterapia. **Objetivo:** Descrever as características clínicas e epidemiológicas da MC em pacientes oncológicos. **Métodos:** Realizou-se um estudo retrospectivo, descritivo e transversal de casos confirmados de MC internados no serviço de neurocirurgia do Hospital de Urgência de Sergipe (Aracaju-Se) durante o período de janeiro de 2011 a agosto de 2012. **Resultados:** Foram estudados 40 pacientes, sendo a média de idade 53,2 anos. A sexta década de vida foi a faixa etária mais acometida. Quanto ao gênero, 24 (60%) feminino e 16 (40%) masculino. O sítio primário mais prevalente da MC foi mama 37,5% dos casos, seguido de pulmão 30%. Os sintomas mais comuns foram cefaleia e distúrbio cognitivo, presentes em 30 (75%) casos. A tomografia computadorizada (TC) foi o método diagnóstico mais utilizado, 36 (90%) pacientes. O tratamento paliativo foi realizado em 25 (62,5%) pacientes, seguido de radioterapia 11 (27,5%), cirurgia 8 (20%) e quimioterapia 3 (7,5%) pacientes. **Conclusão:** A análise clínica e epidemiológica do paciente oncológico é de fundamental importância para o diagnóstico de MC.

Palavras-Chave: Metástase neoplásica; Neoplasia encefálica; Epidemiologia; Manifestações neurológicas

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INTRODUCTION

Brain metastasis (BM) is the most common neurological manifestation of cancer, occurring in 10% to 30% of oncology patients¹ and its incidence has increased in recent decades¹. The primary neoplasms that most frequently metastasize to the central nervous system (CNS) are lung, breast, melanoma, renal, and gastrointestinal cancers². Regarding the location of BM, approximately 80% are found in the cerebral hemispheres, 15% in the cerebellum, and 5% in the brainstem³. Among the signs and symptoms, the most prominent are headache, nausea, vomiting, cognitive dysfunction, seizures, hemiparesis, visual disturbances, and aphasia^{1,3}.

Diagnosis is made using computed tomography (CT) scans and magnetic resonance imaging (MRI) of the head⁴. A biopsy determines the histological type and malignancy grade of the neoplasm⁵. Depending on the number of metastases, lesion location, primary tumor, and patient performance status, specific treatment is indicated⁶. Definitive treatment includes surgery, radiotherapy, and chemotherapy¹. Corticosteroids, anticonvulsants, anticoagulants, antibiotics, and psychotropic drugs are used as symptomatic therapy^{7,8}.

This study aims to characterize the clinical and epidemiological profile of BM to enable early diagnosis and, consequently, choose the appropriate treatment to be instituted, improving quality of life and increasing the survival of cancer patients who develop this complication.

METHODS

This study was a retrospective, cross-sectional, and descriptive field study with a quantitative approach. From January 2011 to August 2012, patients admitted to the Oncology Service at HUSE (Hospital Universitário de Sergipe, Aracaju-SE, Brazil) with clinical and imaging findings of BM were analyzed. The study included patients of both genders and all age groups who had a confirmed diagnosis of BM, agreed to participate in the research, and provided signed informed consent (by the patient or legal guardians).

The evaluated variables included: gender, age, primary tumor, neurological and imaging findings, and survival. The collected data were entered into a computerized database system, and the obtained information was cross-checked and subjected to statistical analysis using the Epi Info software. Simple frequency tables were used for sample characterization.

This study was approved by the Ethics Committee of Hospital Universitário de Sergipe under protocol number CAAE: 6050.0.000.107-10, in compliance with Resolution 196/96 of the Conselho Nacional de Saúde (Brazilian National Health Council).

RESULTS

From January 2011 to August 2012, 40 patients diagnosed with BM were studied. The age range was 26 to 82 years, with a mean age of 53.2 years. The sixth decade of life was the most affected age group. Regarding gender, 24 (60%) were female and 16 (40%) male. In terms of race, 31 (77.5%) were non-white and 9 (22.5%) white (Table 1).

Concerning the number of lesions, 35% were single, and 65% had two or more lesions. Specifically: single lesion (14 patients), two lesions (3 patients, 7.5%), three lesions (3 patients, 7.5%), and four or more lesions (20 patients, 50%).

The most common clinical presentation was neurocognitive disorders and headache, present in 30 (75%) cases, followed by vomiting in 16 (40%) and nausea in 13 (32.5%). Focal manifestations included: hemiparesis (27 patients, 67.5%), seizures (24, 60%), visual disturbances (15, 37.5%), and aphasia (15, 37.5%) (Table 2). The primary sites of BM were breast (15 women, 37%), lung (12 patients, 30%), skin (2, 5%), and prostate, kidney, osteosarcoma, and neuroendocrine tumor (1 case each, 2.5% frequency).

CT was the most used diagnostic method (36 cases, 90%), while MRI was performed in four patients (10%). A biopsy was conducted in five cases (12.5%).

Palliative treatment was the most indicated approach (25 patients, 62.5%). Surgery was performed in eight patients (20%)—with

Table 1. Distribution of patients with neoplasms and central nervous system metastases, according to personal characteristics.

		Number of patients	Percentage (%)
Age (years)	0 – 29	1	2.5
	30 – 39	6	15
	40 – 49	8	20
	50 – 59	10	25
	60 – 69	12	30
	70 – 79	2	5
	≥ 80	1	2.5
Gender	Female	24	60
	Male	16	40
Race	White	9	22.5
	Non-white	31	77.5

curative intent in six cases and to minimize tumor mass effect in the other two. Surgery as the sole treatment was performed in four selected cases. Radiotherapy was administered to 11 patients (27.5%) and chemotherapy to three (7.5%) (Table 2).

The mean survival in our study was 73 days, with a mortality rate of 82.5%. By the end of the study, seven patients (17.5%) remained disease-free after surgery (Table 3).

DISCUSSION

BM represents one of the most significant and feared neurological complications of cancer⁹. The incidence of this condition is increasing due to improved survival rates among cancer patients⁷. Early diagnosis is therefore essential, requiring knowledge of the clinical and epidemiological profile of BM combined with neuroimaging.

The mean age at BM diagnosis in our study was 53.2 years, consistent with other studies reporting an age range of 50-60 years^{10,11}. Regarding gender, there was a female predominance. While Rosen¹² report that BM occurs equally in men and women, disparities emerge when analyzing specific primary tumors.

Lung cancer is the most common primary tumor causing BM^{2,3,9,13}. However, in our study, breast cancer was the leading

primary site (15 women, 37.5%), followed by lung (12 cases, 30%), skin (2, 5%), and prostate, liver, kidney, osteosarcoma and neuroendocrine tumors (1 case each, 2.5%). We attribute this finding to the higher proportion of female patients in our cohort and the rising incidence of breast cancer with subsequent metastatic spread to the CNS. An unknown primary site was identified in 17.5% of cases, exceeding the 15% reported by Bartelt and Lutterbach¹⁴.

Clinical manifestations can be generalized or focal. The most common generalized symptoms were neurocognitive disturbances and headache, followed by vomiting and nausea. The most prevalent focal manifestation was hemiparesis, followed by seizures, visual disturbances, and aphasia, aligning with findings by Christiaans et al.¹⁵ and Soffietti et al.¹⁶.

Headache, the most frequent symptom in BM, occurs in 50% of patients with single metastases¹⁵. It is typically a non-tension-type, lasting ≤10 weeks, and is accompanied by nausea and vomiting. Focal motor deficits and cognitive decline are the most common signs, with the former present in 40% of patients¹⁷. Forsyth and Posner¹⁷ reported nausea and vomiting in 48% of 111 patients, with 66% having BM. Other symptoms include papilledema (25%), gait ataxia (due to posterior fossa, frontal lobe or hydrocephalus involvement), and seizures (25%), which may be focal or generalized.

Diagnosis requires CT and/or MRI, with the latter considered the gold standard^{5,16,18}. On CT, BM typically appears as rounded

Table 2. Distribution of patients with neoplasms and central nervous system metastases, according to clinical characteristics.

	Number of patients	Percentage (%)
A) Clinical presentation		
Headache	30	75
Cognitive impairment	30	75
Hemiparesis	27	67.5
Seizure	24	60
Vomiting	16	40
Aphasia	15	37.5
Visual disturbance	15	37.5
Nausea	13	32.5
B) Primary tumor site		
Breast	15	37.5
Lung	12	30
Undetermined	7	17.5
Skin	2	5
Prostate	1	2.5
Kidney	1	2.5
Osteosarcoma	1	2.5
Neuroendocrine	1	2.5
C) Diagnostic method		
Computed tomography (CT)	36	90
Biopsy	5	12.5
Magnetic resonance imaging (MRI)	4	10
D) Treatment		
Palliative care	25	62.5
Radiotherapy	11	27.5
Surgery	8	20
Chemotherapy	3	7.5

Table 3. Distribution of patients with neoplasms and central nervous system metastases, according to survival after diagnosis.

Survival after diagnosis of metastases (days)	Number of patients	Percentage (%)
0–30	12	30
30–60	8	20
60–90	9	22.5
90–120	1	2.5
120–150	1	2.5
150–180	1	2.5
180–365	4	10
>365	4	10

lesions with diffuse or ring-like enhancement, surrounded by disproportionate perilesional edema³. On MRI, metastases are iso- or hypointense on T1 and iso- or hyperintense on T2, surrounded by vasogenic edema¹⁹. In our study, CT was used in 36 patients (90%) and MRI in four (10%), consistent with prior research¹⁸. Biopsy helps differentiate primary from metastatic lesions and may suggest the primary tumor site²⁰. In our cohort, biopsy confirmed BM in five patients (12.5%).

Regarding treatment, surgery aims to establish diagnosis and relieve mass effect²¹. It is indicated when the primary tumor is controlled, brain lesions are accessible, and the number of metastases does not exceed three¹⁹. In emergencies, surgery may prevent fatal complications^{22,23}. Radiotherapy combined with surgery improves survival more than radiotherapy alone²⁴. Chemotherapy has limited efficacy due to poor blood-brain barrier penetration³. In our study, palliative treatment was administered to 25 patients (62.5%). Surgery was performed in eight cases (20%) - six with curative intent and two to reduce mass effect. Radiotherapy was given to 11 patients (27.5%) and chemotherapy to three (7.5%).

Post-BM diagnosis survival is typically limited to 3-9 months^{3,24}. In our study, mean survival was 73 days with a mortality rate of 82.5%. Disease-free survival after surgery was observed in seven patients (17.5%).

Since all analyzed patients exhibited neurological symptoms before BM diagnosis, cancer patients presenting with generalized or focal neurological manifestations should undergo neuroimaging for early BM detection and prompt treatment initiation.

CONCLUSION

The clinical and epidemiological profile is of great value for the diagnosis of BM. Our patients exhibited some symptoms prior to imaging diagnosis. Among the most prevalent were neurocognitive disorders, headache, and hemiparesis. Notably, the average age and survival were lower than those reported in the literature, there was a higher incidence of BM in females, and breast cancer was the most frequent primary tumor associated with BM. The most commonly used diagnostic

method was CT, followed by MRI. Most patients underwent palliative treatment.

REFERENCES

1. Kanner AA, Bokstein F, Blumenthal DT, Ram Z. Surgical therapies in brain metastasis. *Semin Oncol*. 2007;34(3):197-205. <http://doi.org/10.1053/j.seminoncol.2007.03.011>. PMID:17560981.
2. Barnholtz-Sloan JS, Sloan AE, Davis FG, Vignea FD, Lai P, Sawaya RE. Incidence proportions of brain metastases in patients diagnosed (1973 to 2001) in the Metropolitan Detroit Cancer Surveillance System. *J Clin Oncol*. 2004;22(14):2865-72. <http://doi.org/10.1200/JCO.2004.12.149>. PMID:15254054.
3. Santos AJ, Franco CMR, Borges LRR, Malheiros SMF, Gabbai AA. Metástases cerebrais. *Rev Neurociencias*. 2001;9(1):20-6. <http://doi.org/10.34024/rnc.2001.v9.8929>.
4. Najeeb AA, Amiri NA, Miola UJ, Moutaery KA, Ostertag CB. Brain metastases: surgery, radiosurgery, and whole-brain radiation therapy. *Pan Arab J Neurosurg*. 2008;12:19-27.
5. Davis P, Hudgins PA, Peterman SB, Hoffman JC Jr. Diagnosis of cerebral metastases: double-dose delayed CT vs contrast enhanced MR imaging. *AJNR Am J Neuroradiol*. 1991;12(2):293-300. PMID:1902031.
6. Bajard A, Westeel V, Dubiez A, et al. Multivariate analysis of factors predictive of brain metastases in localised non-small cell lung carcinoma. *Lung Cancer*. 2004;45(3):317-23. <http://doi.org/10.1016/j.lungcan.2004.01.025>. PMID:15301872.
7. Rashmi K, Dubey A, Vijay K, Eisenstat DD. Use of corticosteroids in patients with brain metastasis. *Oncol Exchange*. 2010;9:7-10.
8. Westphal M, Heese O, Wit DM. Intracranial metastases: therapeutic options. *Ann Oncol*. 2003;14(Suppl 3):4-10. <http://doi.org/10.1093/annonc/mdg741>. PMID:12821532.
9. Fabi A, Felici A, Metro G, et al. Brain metastases from solid tumors: disease outcome according to type of treatment and therapeutic resources of the treating center. *J Exp Clin Cancer Res*. 2011;30(1):10. <http://doi.org/10.1186/1756-9966-30-10>. PMID:21244695.
10. Selek U, Chang EL, Hassenbusch SJ 3rd, et al. Stereotactic radiosurgical treatment in 103 patients for 153 cerebral melanoma metastases. *Int J Radiat Oncol Biol Phys*. 2004;59(4):1097-106. <http://doi.org/10.1016/j.ijrobp.2003.12.037>. PMID:15234044.
11. Fife KM, Colman MH, Stevens GN, et al. Determinants of outcome in melanoma patients with cerebral metastases. *J Clin Oncol*. 2004;22(7):1293-300. <http://doi.org/10.1200/JCO.2004.08.140>. PMID:15051777.
12. Rosen ST. Brain metastases. New York: Springer; 2007. 4 p.

13. Gavrilovic IT, Posner JB. Brain metastases: epidemiology and pathophysiology. *J Neurooncol.* 2005;75(1):5-14. <http://doi.org/10.1007/s11060-004-8093-6>. PMID:16215811.
14. Bartelt S, Lutterbach J. Brain metastases in patients with cancer of unknown primary. *J Neurooncol.* 2003;64(3):249-53. <http://doi.org/10.1023/A:1025621819250>. PMID:14558600.
15. Christiaans MH, Kelder JC, Arnoldus EPJ, Tijssen CC. Prediction of intracranial metastases in cancer patients with headache. *Cancer.* 2002;94(7):2063-8. <http://doi.org/10.1002/cncr.10379>. PMID:11932910.
16. Soffietti R, Cornu P, Delattre JY, et al. EFNS Guidelines on diagnosis and treatment of brain metastases: report of an EFNS Task Force. *Eur J Neurol.* 2006;13(7):674-81. <http://doi.org/10.1111/j.1468-1331.2006.01506.x>. PMID:16834697.
17. Forsyth PA, Posner JB. Headaches in patients with brain tumors: a study of 111 patients. *Neurology.* 1993;43(9):1678-83. <http://doi.org/10.1212/WNL.43.9.1678>. PMID:8414011.
18. Lalondrelle S, Khoo V. Brain metastases. *BMJ Clin Evid.* 2009;2009:1018. PMID:19445757.
19. Schackert G. Surgery of brain metastases: pro and contra. *Onkologie.* 2002;25(5):480-1. PMID:12415204.
20. Takahashi JA, Llena JF, Hirano A. Pathology of cerebral metastases. *Neurosurg Clin N Am.* 1996;7(3):345-67. [http://doi.org/10.1016/S1042-3680\(18\)30366-8](http://doi.org/10.1016/S1042-3680(18)30366-8). PMID:8823768.
21. Onishi FJ, Melo JGSP, Melo PMP, Lanzoni OP, Settanni F, Ferraz FAP. Tratamento cirúrgico de metástases intracranianas. *Rev Neurociências.* 2005;13(1):11-6. <http://doi.org/10.34024/rnc.2005.v13.8839>.
22. Sheehan J, Niranjana A, Flickinger JC, Kondziolka D, Lunsford LD. The expanding role of neurosurgeons in the management of brain metastases. *Surg Neurol.* 2004;62(1):32-40. <http://doi.org/10.1016/j.surneu.2003.10.033>. PMID:15226065.
23. Bradley KA, Mehta MP. Management of brain metastases. *Semin Oncol.* 2004;31(5):693-701. <http://doi.org/10.1053/j.seminoncol.2004.07.012>. PMID:15497123.
24. Patchell RA, Tibbs PA, Walsh JW, et al. A randomized trial of surgery in the treatment of single metastases to the brain. *N Engl J Med.* 1990;322(8):494-500. <http://doi.org/10.1056/NEJM199002223220802>. PMID:2405271.

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Efficacy and Safety of Unilateral Biportal Endoscopy Compared to Microscopic and Uniportal Approaches in Lumbar Spinal Stenosis: evidence from 25 studies

Eficácia e Segurança da Endoscopia Biportal Unilateral em Comparação com Abordagens Microscópicas e Uniportais na Estenose Lombar: evidências de 25 estudos

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Sérgio Cavalheiro¹ 

ABSTRACT

Background: Lumbar spinal stenosis (LSS) is a prevalent degenerative disorder in the elderly, frequently necessitating surgical decompression. Microscopic decompression has long been established, but minimally invasive approaches, including monoportal and unilateral biportal endoscopy (UBE), are increasingly utilized for their potential to limit tissue damage and enhance recovery. This systematic review and meta-analysis compared the efficacy and safety of UBE with monoportal and microscopic techniques. **Methods:** Eligible studies compared at least two techniques — UBE, monoportal endoscopy, or microscopic decompression — for LSS. Outcomes assessed included Oswestry Disability Index (ODI), Visual Analog Scale (VAS) for pain, intraoperative blood loss, operative time, hospital stay, patient satisfaction, reoperation rates, and complications (dural tears, infections, hematomas, neural injury). Protocol: PROSPERO (CRD420251079604). **Results:** Twenty-five studies with 3,496 patients were included. Compared with microscopic surgery, UBE yielded greater ODI improvement ($p = 0.00001$), less blood loss, shorter hospital stays, and higher satisfaction ($p = 0.004$). Versus monoportal endoscopy, UBE showed similar ODI, VAS, operative time, and satisfaction, but greater blood loss and slightly longer hospitalization. **Conclusions:** UBE provides outcomes equal to or superior to monoportal and microscopic decompression with comparable safety. Nonetheless, evidence may reflect publication or institutional bias, highlighting the need for high-quality randomized trials with transparent conflict-of-interest reporting.

Keywords: Lumbar spinal stenosis; Unilateral biportal endoscopy; Endoscopic spine surgery; Microscopic decompression; Minimally invasive spine surgery; Systematic review

RESUMO

Introdução: A estenose lombar (LSS) é uma condição degenerativa comum em idosos, frequentemente exigindo descompressão cirúrgica. Abordagens minimamente invasivas endoscópicas têm recebido atenção pelo potencial de reduzir o dano aos tecidos moles e melhorar a recuperação. **Métodos:** Foram incluídos estudos comparativos: UBE, endoscopia uniportal e descompressão microscópica para o tratamento da estenose lombar. Os desfechos de melhora clínica e complicações foram analisados. **Resultados:** Foram incluídos 25 estudos, totalizando 3.496 pacientes. A UBE apresentou melhora significativamente maior no ODI ($p = 0,00001$), menor perda sanguínea intraoperatória e menor tempo de internação. A satisfação dos pacientes também foi maior no grupo UBE em relação à cirurgia microscópica ($p = 0,004$). Quando comparada à endoscopia uniportal, a UBE apresentou resultados semelhantes em ODI, EVA, tempo cirúrgico e satisfação. **Conclusões:** A UBE proporciona resultados clínicos semelhantes ou superiores à endoscopia uniportal e à descompressão microscópica em termos de melhora funcional, mantendo perfil de segurança equivalente. Contudo, considerando a

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relativa novidade da UBE e a concentração de estudos favoráveis em centros de alto volume, deve-se ponderar a possibilidade de viés de publicação e comercial.

Palavras-Chave: *Estenose lombar; Endoscopia biportal unilateral; Cirurgia endoscópica da coluna; Descompressão microscópica; Cirurgia minimamente invasiva da coluna; Revisão sistemática*

INTRODUCTION

Lumbar spinal stenosis (LSS) is a common degenerative condition affecting the aging population, characterized by a progressive narrowing of the spinal canal that leads to neural compression, back pain, neurogenic claudication, and functional limitations. Surgical decompression remains the cornerstone of treatment for patients unresponsive to conservative measures, with traditional microscopic decompression being a widely accepted standard. However, conventional techniques, including open and microscopic approaches, are associated with iatrogenic muscle and soft-tissue injury, prolonged recovery, and potential postoperative back pain due to muscle denervation and facet joint disruption.

In response to the limitations of traditional techniques, minimally invasive spinal surgery has evolved significantly over the past two decades. Among these innovations, UBE — first introduced in the early 2010s and largely refined in East Asia — has gained traction as a promising alternative for lumbar decompression. The UBE technique utilizes two independent portals: one for endoscopic visualization and another for the working instruments, thus allowing simultaneous bimanual manipulation similar to open surgery while preserving the benefits of endoscopy. This dual-portal system offers superior visualization, continuous saline irrigation to maintain a clear field, and the flexibility to use conventional surgical tools, reducing the learning curve and the need for specialized equipment^{1,2}.

Early studies and recent meta-analyses have demonstrated that UBE achieves comparable or superior clinical outcomes to both microendoscopic decompression and uniportal endoscopy (UE), especially in terms of short-term pain relief, functional improvement, and reduced muscle trauma³⁻⁵. Moreover, UBE is associated with a lower rate of extensive facet joint destruction, enabling better preservation of spinal stability^{2,6}. Nonetheless, as with any surgical innovation, concerns remain regarding

complication profiles, particularly during the initial learning curve. Reported complications include dural tears, epidural hematomas, incomplete decompression, transient neurological deficits, and postoperative instability, with an estimated mean complication rate of 5%–8%^{7,8}.

Given the rapid global adoption of UBE and the growing body of literature supporting its efficacy, a comprehensive evaluation of both its clinical benefits and safety profile is essential. This systematic review aims to critically assess the comparative effectiveness of UBE against MONO and microscopic decompression in the treatment of LSS. Particular attention will be paid to operative time (OT), postoperative recovery, extent of decompression, and complication rates to guide evidence-based surgical decision-making in the era of minimally invasive spine surgery.

METHODS

This systematic review was conducted following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA 2020 - Figure 1) guidelines to provide a comprehensive and methodologically rigorous comparison between UBE, monoportal endoscopic surgery, and microscopic decompression for the treatment of lumbar spinal stenosis. The review protocol was established before the initiation of the study, ensuring a structured and unbiased approach to study selection, data extraction, and analysis. The protocol for this systematic review was prospectively registered in the International Prospective Register of Systematic Reviews (PROSPERO) under registration number CRD420251079604.

A comprehensive literature search was performed across three major databases: PubMed (MEDLINE), Embase, and the Cochrane Central Register of Controlled Trials (CENTRAL). To maximize the sensitivity of the search and ensure broad coverage of the

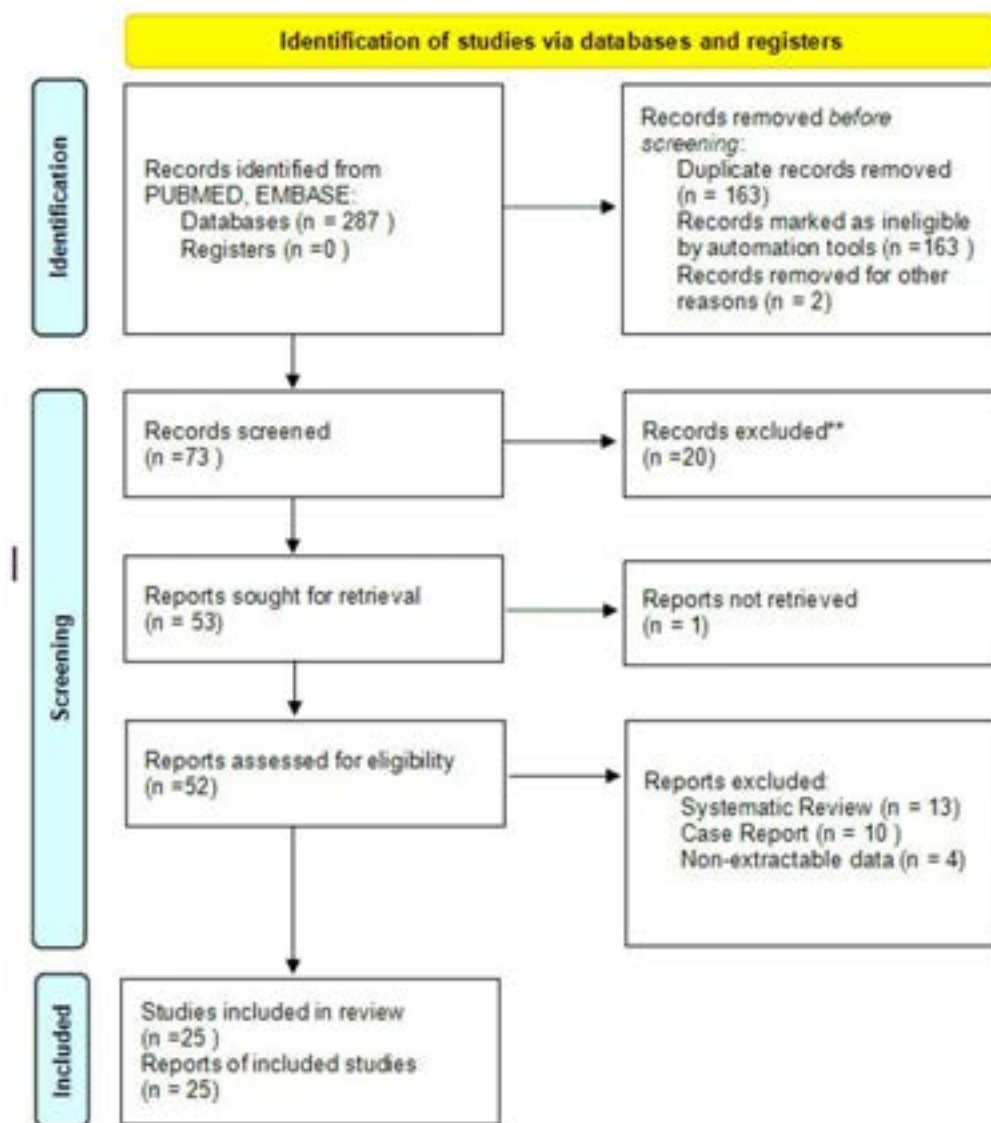


Figure 1. PRISMA flow diagram.

available literature, no restrictions were applied regarding language or date of publication. The search strategy was constructed using a combination of Medical Subject Headings (MeSH) and free-text terms related to “lumbar spinal stenosis,” “unilateral biportal endoscopy,” “monoportal endoscopy,” and “microscopic decompression,” and adapted appropriately for each database.

Eligibility criteria were defined based on the PICOS framework, including studies that directly compared at least two of the surgical techniques under investigation—UBE, monoportal endoscopy, or microscopic surgery—for patients diagnosed with lumbar spinal stenosis. Both randomized controlled trials (RCT) and non-randomized

comparative studies (prospective or retrospective) were included. Case series without a comparator, reviews, letters to the editor, conference abstracts, and animal or cadaveric studies were excluded.

Two independent reviewers screened titles and abstracts for potential inclusion. Studies that met initial criteria were retrieved in full and assessed independently by the same reviewers for final inclusion, with any disagreements resolved through discussion or arbitration by a third reviewer. Data were then extracted using a standardized form developed specifically for this review. Extracted data included study characteristics (authors, year, country, and design), patient demographics, surgical technique details, and clinical outcomes

such as VAS for leg and back pain, ODI, OT, length of hospital stay, complication rates, reoperation rates, and other patient-reported outcome measures (PROMs). Data extraction was performed independently by two reviewers and cross-checked to ensure accuracy.

The risk of bias of the included studies was assessed using validated tools appropriate for each study design. For RCTs, the Cochrane RoB 2.0 tool (Risk of Bias 2.0 -Cochrane tool for RCTs) was applied, evaluating domains such as the randomization process, deviations from intended interventions, missing outcome data, outcome measurement, and selective reporting. For non-randomized studies, the Risk of Bias In Non-randomized Studies of Interventions (ROBINS-I) tool was used, examining potential confounding, selection bias, classification of interventions, and outcome assessment. Disagreements in bias assessment were resolved through discussion or third-party adjudication.

The results were synthesized qualitatively and, where data were sufficiently homogeneous, a quantitative meta-analysis was planned. When applicable, pooled analyses would be conducted using a random-effects model to account for expected clinical variability. For continuous outcomes, mean differences (MD) and standard deviations (SD) would be used, while dichotomous outcomes would be summarized using risk ratios (RR) or odds ratios (OR) with corresponding 95% confidence intervals (CI). Statistical heterogeneity would be assessed using the I^2 statistic, with values above 50% interpreted as indicating substantial heterogeneity.

In cases where at least ten studies contributed to a pooled estimate for a given outcome, potential publication bias would be explored using funnel plots and Egger's test.

RESULTS

A systematic search using predefined eligibility criteria yielded 25 studies for data synthesis⁹⁻³³, including 3 RCTs; the remaining studies consisted of prospective or retrospective comparative designs. Regarding the geographic distribution of the studies, the majority originated from Eastern countries, with 11 from China, 10 from South Korea, and one each from Germany, the United States, and Japan.

A total of 3,496 patients were analyzed in the final synthesis, with a mean age of 65.7 years and a SD of 6.51. Seventeen studies reported a follow-up period equal to or longer than one year.

Risk of bias analysis of RCTs (Figure 2)



Figure 2. ROB 2 of 3 Randomized controlled trials (RCT).

Risk of bias of non-RCTs (Figure 3)



Figure 3. ROBINS-I of 22 non-RCTs.

The results of the meta-analysis will be presented below according to the predefined outcomes. These include functional outcomes measured by the ODI, pain scores assessed using the VAS for both back and leg pain, intraoperative blood loss, OT, LOS, patient satisfaction with surgery, and incidence of secondary surgeries. Additionally, recurrence of symptoms, overall complication rates, and specific adverse events, including dural tears, surgical site infections, epidural hematomas, and neural injuries, were also evaluated.

ODI

Ten studies compared preoperative and postoperative ODI scores between UBE and uniportal endoscopic techniques.

No significant difference was observed in the magnitude of ODI improvement between the two approaches ($p = 0.68$), as seen in Figure 4.

Eleven studies compared UBE with microscopic surgery. The magnitude of ODI improvement was significantly greater in the UBE group compared to the microscopic group (Figure 5) ($p = 0.00001$).

VAS (axial)

Nine studies compared axial VAS scores between UBE and uniportal endoscopic techniques. No significant difference was observed between the two groups (Figure 6).

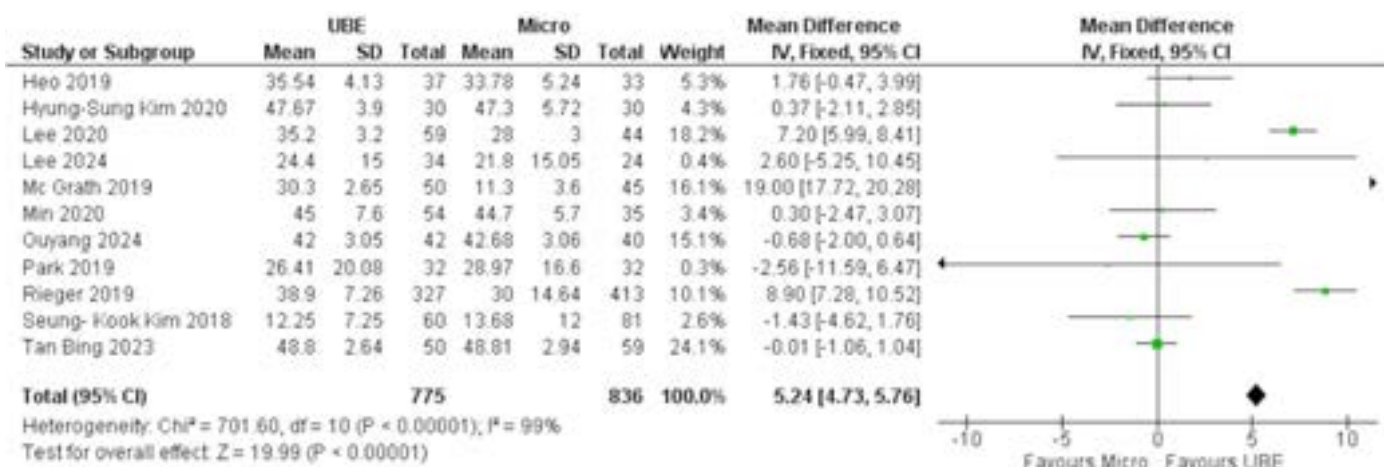


Figure 4. UBE vs Monoportal showing no difference in ODI scores.

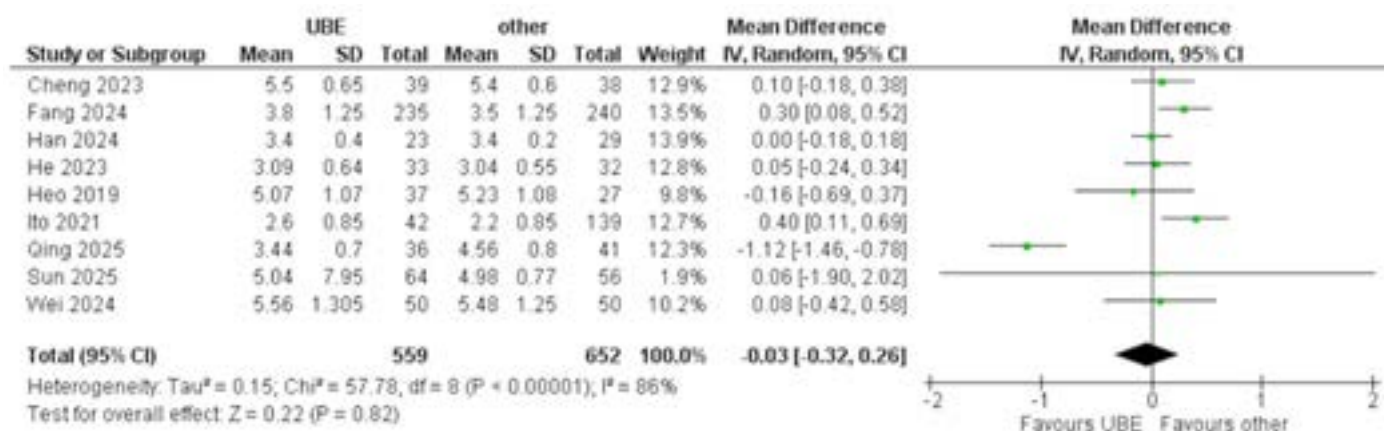


Figure 5. UBE had better ODI scores compared to microscopic decompression.

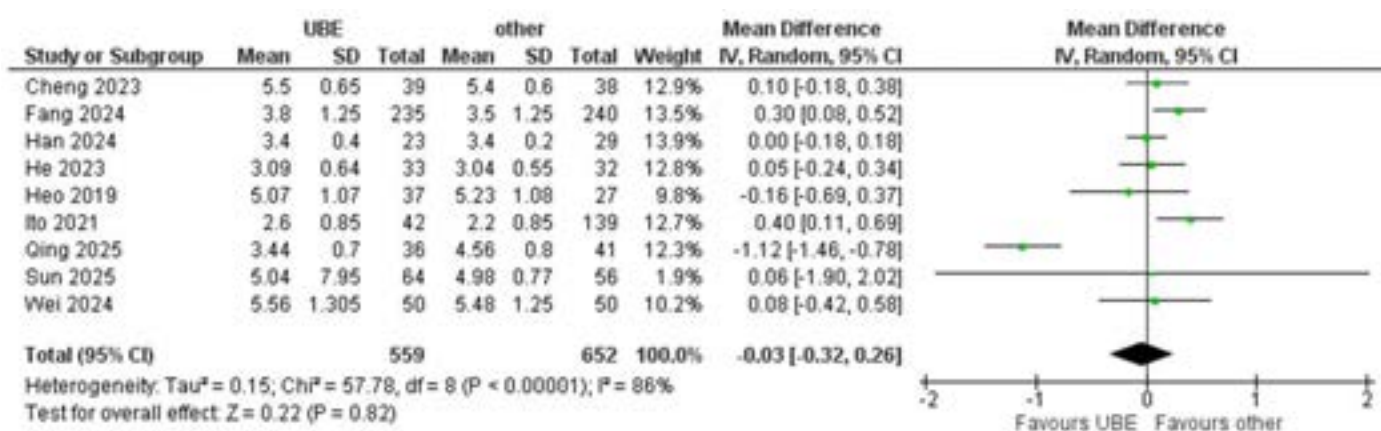


Figure 6. UBE and monoportal had the same axial VAS results.

Similarly, in the comparison between UBE and microscopic surgery across 11 studies, no significant difference was observed in axial VAS scores as seen in Figure 7 ($p = 0.19$).

VAS (leg)

Eight studies compared leg VAS scores between UBE and uniportal endoscopic techniques (Figure 8), showing no significant difference in outcomes ($p = 0.72$).

Eight studies also compared leg VAS scores between UBE and microscopic surgery, without demonstrating any statistically significant difference between the groups (Figure 9).

Intraoperative blood loss

An interesting finding emerged from the analysis of intraoperative blood loss. When compared to the uniportal endoscopic technique, UBE was associated with a higher volume of bleeding. However, when compared to the microscopic technique, UBE demonstrated a more favorable bleeding profile, with a lower mean blood loss (Figures 10 and 11).

Operative time

Regarding operative time, UBE demonstrated comparable durations when analyzed against both the uniportal technique ($p = 0.1$) and microscopic surgery ($p = 0.2$).

Length of hospital stay

Regarding the LOS, a shorter duration was observed for the uniportal endoscopic technique compared to UBE. However,

when UBE was compared to microscopic surgery, it was associated with a significantly shorter hospital stay (Figure 12).

Patient satisfaction

Patient satisfaction with surgery was subjectively reported as high across studies. When comparing the two endoscopic approaches—biportal and uniportal—no significant differences in satisfaction levels were observed. However, in comparisons between UBE and microscopic surgery, patients in the UBE group reported a higher degree of satisfaction with the surgical outcome ($p=0.004$) (Figure 13).

Incidence of secondary surgeries

Reoperations were analyzed and found to be rare events overall. When comparing the reoperation rates at the index level between UBE and both monoportal and microscopic techniques, no statistically significant differences were observed.

Dural tears

No significant differences were found in the incidence of dural tears when comparing UBE with either the uniportal endoscopic technique ($p=0.76$) or microscopic surgery ($p=0.36$).

Surgical site infections

The incidence of postoperative infections was low across studies, and no significant differences were observed between UBE and either the uniportal ($p=0.68$) or microscopic techniques ($p=0.26$).

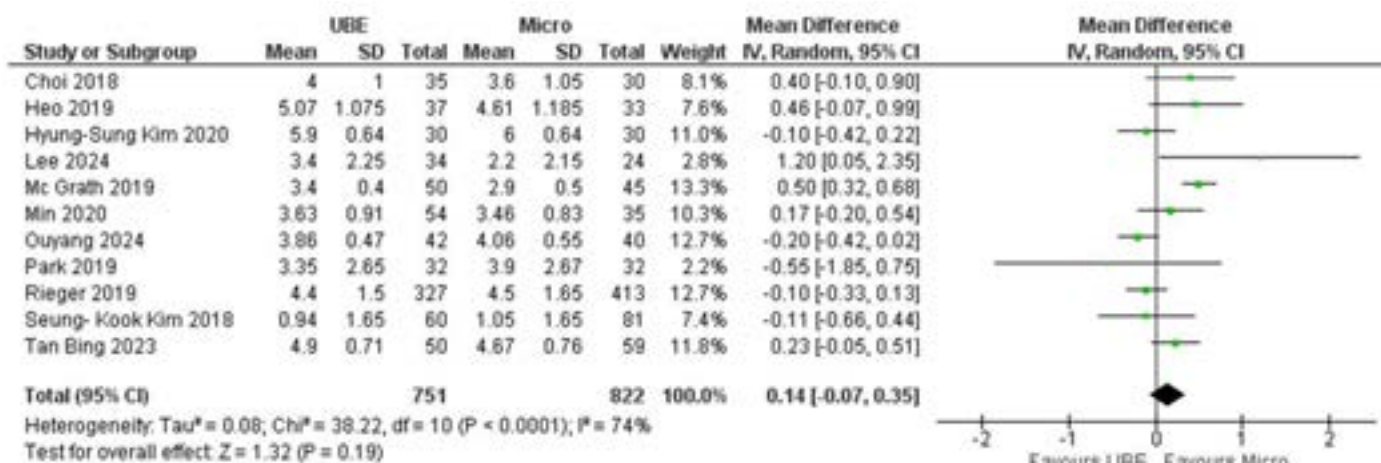


Figure 7. UBE and microscopic had the same axial VAS results.

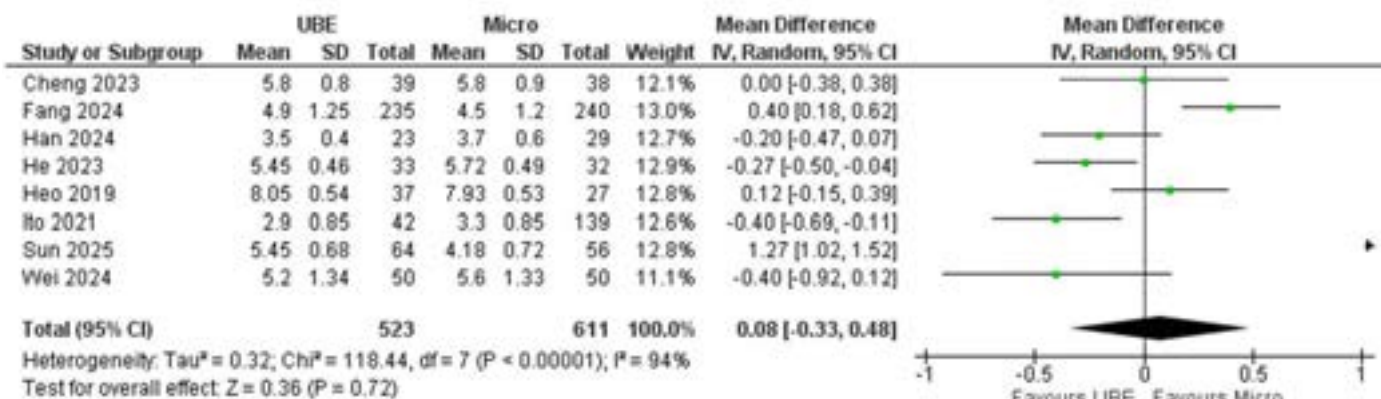


Figure 8. UBE and monoportal had the same leg VAS results.

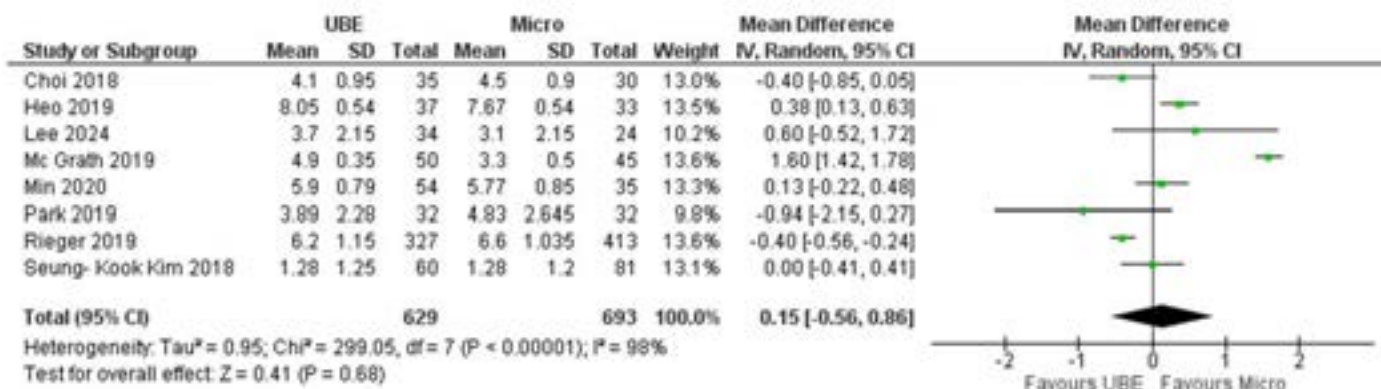


Figure 9. UBE and microscopic had the same leg VAS results.

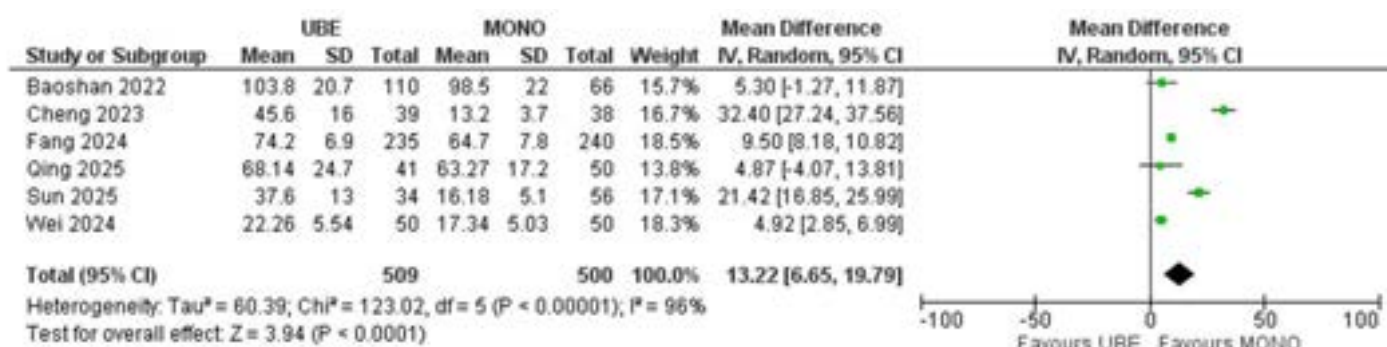


Figure 10. Monoportal had less blood loss than UBE.

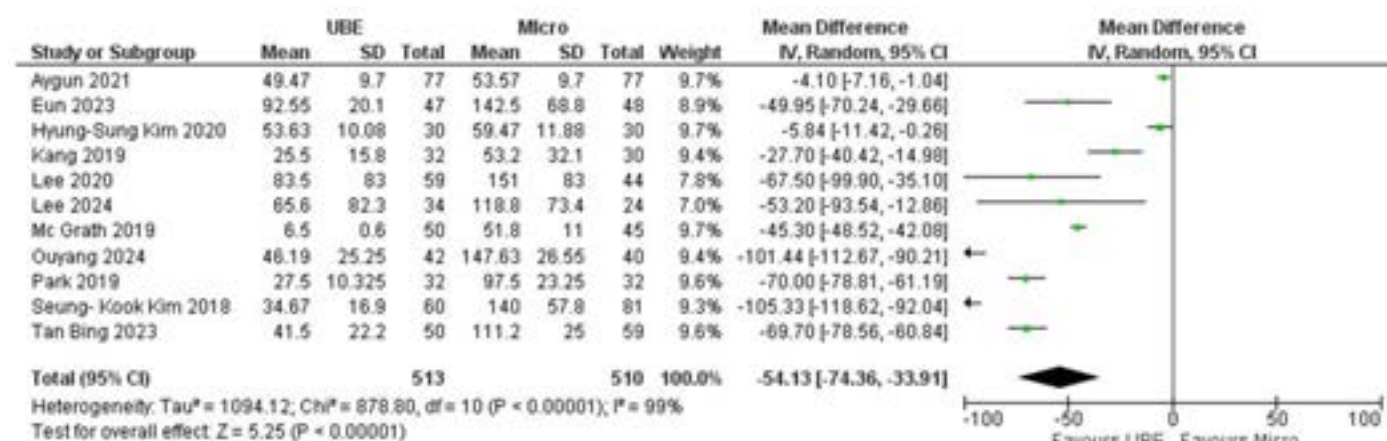


Figure 11. UBE had less blood loss than microscopic decompression.

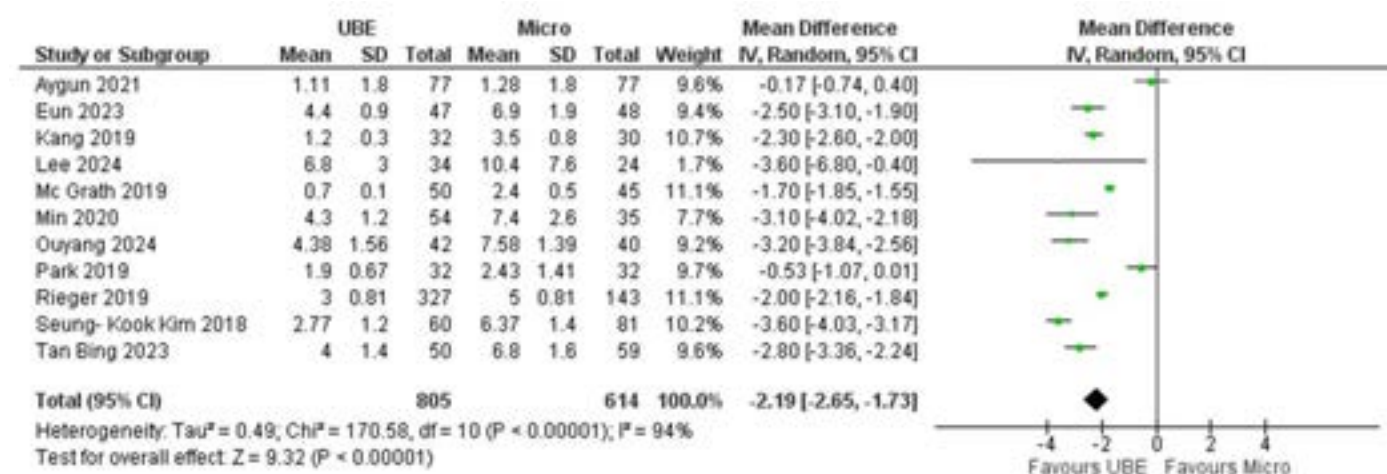


Figure 12. UBE had a shorter LOS than Monoportal.

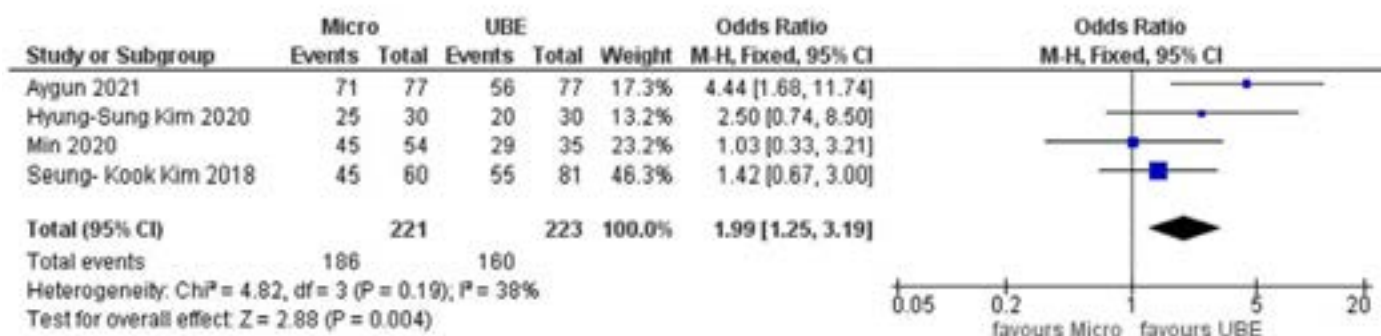


Figure 13. Patient satisfaction was higher in UBE patients than Microsurgery patients.

Epidural hematomas

The occurrence of epidural hematoma was rare and showed no statistically significant difference when comparing UBE to uniportal ($p=0.37$) or microscopic approaches ($p=0.42$).

Neural injuries

Only the study by Fang et al. (2024) specifically evaluated and compared the incidence of neurological injury between UBE and the uniportal endoscopic technique. Rates of neural injury were low and comparable among the three methods, with no significant differences between UBE, uniportal, and microscopic surgery.

SYNTHESIS OF OUTCOMES

UBE demonstrated superior outcomes in several domains when compared to microscopic surgery. Functional improvement, measured by the ODI, was significantly greater in the UBE group. UBE was also associated with lower intraoperative blood loss than microscopic surgery and a shorter hospital stay. Additionally, patients who underwent UBE reported higher satisfaction levels. When compared to UE, UBE achieved similar improvements in ODI and VAS scores, with no significant differences in OT or complication rates.

MONO was associated with the shortest hospital stay among the three techniques and resulted in less intraoperative bleeding than UBE, reflecting its highly minimally invasive nature. However, it did not show superiority over UBE or microscopic surgery in functional or pain-related outcomes.

Microscopic surgery did not demonstrate superior results in any of the analyzed outcomes. OT, reoperation rates, and complication profiles—including dural tears, infections, epidural hematomas,

and neural injuries—were comparable across all three approaches (Table 1).

DISCUSSION

This systematic review provides an updated synthesis of the evidence comparing UBE with MONO and microscopic decompression for LSS. Our findings highlight UBE as a viable and increasingly utilized minimally invasive surgical option, offering a balance of operative efficiency, clinical effectiveness, and acceptable complication rates.

One of the most consistent findings across the reviewed studies is the operative efficiency of UBE. The dual-portal system permits independent control of the endoscope and working instruments, thereby improving visualization and facilitating bimanual surgical techniques. This flexibility overcomes some of the ergonomic constraints inherent in MONO, where both visualization and instrument manipulation are limited to a single working channel^{1,15}. Several meta-analyses and comparative studies support that UBE is associated with significantly shorter OTs, particularly in retrospective cohorts^{3,5}. Although these benefits appear more pronounced in retrospective analyses than in prospective studies, the trend toward reduced operative duration with UBE remains consistent.

Initial concerns regarding muscle trauma due to the second incision in UBE have not been substantiated by recent data. On the contrary, multiple studies demonstrate that UBE induces no

Table 1. Summary of the studied outcomes.

Outcome	UBE	MONO	MIC
ODI improvement	✓	✓	X
VAS (back pain)	✓	✓	✓
VAS (leg pain)	✓	✓	✓
Intraoperative blood loss	X	✓	X
Operative time	✓	✓	✓
Length of hospital stay	X	✓	X
Patient satisfaction	✓	✓	X
Reoperation rate	✓	✓	✓
Dural tears	✓	✓	✓
Surgical site infection (SSI)	✓	✓	✓
Epidural hematoma	✓	✓	✓
Neural injuries	✓	✓	✓

UBE = unilateral biportal endoscopy; MONO= monoportal endoscopy, MIC = Microsurgical or tubular decompression.

greater, and possibly even less, paraspinal muscle damage than monoportal or microscopic techniques. The UBE approach employs a muscle-splitting trajectory, which preserves muscle integrity and the posterior ligamentous complex, in contrast to the stripping and retraction required in open or microscopic procedures^{8,34}. Biochemical markers of tissue injury (e.g., CPK, CRP) and postoperative imaging have shown lower muscle disruption and a good preservation of anatomical structures with UBE³⁵.

When comparing clinical outcomes, UBE is consistently associated with similar or superior improvements in pain and disability, as measured by VAS and ODI scores. Studies comparing UBE to microscopic decompression have reported equivalent or better early postoperative pain control, lower opioid requirements, and shorter hospital stays^{4,7,16}. In addition, UBE achieves comparable or greater radiological decompression, with some studies documenting a larger increase in postoperative dural sac area and improved facet joint preservation^{6,18,21,34,36-38}.

Importantly, the safety profile of UBE appears acceptable, with most studies reporting overall complication rates between 5% and 8%^{5,7,8,39}.

Common complications include dural tears, epidural hematomas, transient neurological deficits, and—in rare cases—postoperative instability. Notably, these events are more prevalent during the early learning curve, suggesting that surgical experience plays a significant

role in minimizing risk^{7,8,40,41}. The dual-port setup may also enhance the surgeon's ability to achieve hemostasis and avoid inadvertent injury due to its improved visualization and working angle.

Despite promising results for UBE and monoportal techniques, several limitations in the current evidence base must be acknowledged. Most included studies are retrospective, increasing susceptibility to selection bias and incomplete reporting of complications. The literature also reveals a tendency toward publication bias, with favorable outcomes—particularly from novel techniques like UBE—being more likely to appear in print. In addition, industry influence and narrative bias are not uncommon, with nearly half of spinal surgery reviews showing some form of “spin” in conclusions. Transparent reporting of conflicts of interest is essential, especially given the proprietary nature of UBE instrumentation and its commercial promotion.

MONO offers advantages such as reduced soft tissue damage and faster recovery. However, its delicate instrumentation can pose challenges in elderly patients and multilevel stenosis, where longer surgeries may increase risk. Interestingly, our review did not show increased OTs, possibly due to selection bias, as more complex cases may have been treated by other approaches. Similarly, while symptomatic epidural hematomas are often suspected to occur more frequently after endoscopic surgery, this was not observed in the reviewed data, perhaps due to improved

hemostatic technique or underreporting, especially as such complications are rarely mentioned even in microscopic series.

When comparing techniques, both UBE and MONO demonstrate perioperative advantages over conventional microscopic surgery, including less postoperative pain, shorter hospital stays, and quicker return to function. Still, the long-term outcomes, particularly for back and leg pain, tend to equalize across techniques when adequate decompression is achieved. Although microsurgery had worse outcomes in some perioperative metrics, it remains the gold standard in certain scenarios. It allows superior visualization of anatomical planes with a 3D view of the surgical field, facilitates more reliable dural repair in cases of cerebrospinal fluid leak, and is supported by the longest clinical follow-up in the literature.

CONCLUSION

This systematic review provides a comprehensive comparison of UBE, monoportal endoscopy, and microscopic decompression for the treatment of lumbar spinal stenosis, incorporating data from 25 studies and nearly 3,500 patients. The findings indicate that UBE offers clinical outcomes that are at least equivalent—and in some cases superior—to those achieved with established techniques. In particular, UBE was associated with greater functional improvement compared to microscopic surgery, as well as shorter hospital stays and higher patient satisfaction. Although MONO demonstrated advantages in terms of reduced intraoperative bleeding and the shortest length of stay, its overall efficacy was comparable to that of UBE. Notably, microscopic decompression did not show superiority in any assessed outcome, despite its long-standing use and surgical familiarity.

All three approaches demonstrated similarly low rates of complications and reoperations, confirming that, when performed by experienced surgeons, each technique can be considered safe and effective. However, the relative novelty of UBE, the geographic concentration of studies in regions with strong endoscopic training programs, and the potential influence of device manufacturers highlight the importance of interpreting these results with caution.

Future research should prioritize high-quality, multicenter RCTs with standardized outcome reporting. In particular, extended

follow-up is essential to determine whether the initial advantages observed with UBE translate into sustained long-term benefits, as only prolonged observation can truly establish the superiority of one technique over another. Until such evidence is available, the choice of surgical approach should remain individualized, based on the surgeon's expertise, the patient's profile, and institutional capabilities.

REFERENCES

1. Park DK, Weng C, Zakko P, Choi DJ. Unilateral biportal endoscopy for lumbar spinal stenosis and lumbar disc herniation. *JBJS Essent Surg Tech*. 2023;13(2). <http://doi.org/10.2106/JBJS.ST.22.00020>. PMID:38274147.
2. Pao JL. A review of unilateral biportal endoscopic decompression for degenerative lumbar canal stenosis. *Int J Spine Surg*. 2021;15(Suppl 3):S65-71. <http://doi.org/10.14444/8165>. PMID:35027470.
3. Meng H, Su N, Lin J, Fei Q. Comparative efficacy of unilateral biportal endoscopy and micro-endoscopic discectomy in the treatment of degenerative lumbar spinal stenosis: a systematic review and meta-analysis. *J Orthop Surg Res*. 2023;18(1):814. <http://doi.org/10.1186/s13018-023-04322-2>. PMID:37907922.
4. Zhuang HX, Guo SJ, Meng H, Lin JS, Yang Y, Fei Q. Unilateral biportal endoscopic spine surgery for lumbar spinal stenosis: a systematic review and meta-analysis. *Eur Rev Med Pharmacol Sci*. 2023;27(11):4998-5012. http://doi.org/10.26355/eurrev_202306_32617. PMID:37318474.
5. Li K, Zhang Z, Ran J, Ma L, Meng X. Unilateral Endoscopic and Unilateral Biportal Endoscopic surgery for lumbar spinal stenosis: a systematic review and meta-analysis. *Front Surg*. 2025;12:1585783. <http://doi.org/10.3389/fsurg.2025.1585783>. PMID:40589530.
6. Dou C, Yu Q, Zhang W, Ma L, Meng X. Comparison of Biportal Endoscopic Technique and Conventional Unilateral Laminectomy for Bilateral Decompression (ULBD) for multi-level degenerative lumbar spinal stenosis in elderly people. *Orthop Surg*. 2025;17(8):2302-12. <http://doi.org/10.1111/os.70084>. PMID:40528425.
7. Chen Z, Zhou H, Wang X, Liu Z, Liu W, Luo J. Complications of unilateral biportal endoscopic spinal surgery for lumbar spinal stenosis: a meta-analysis and systematic review. *World Neurosurg*. 2023;170:e371-9. <http://doi.org/10.1016/j.wneu.2022.11.019>. PMID:36368457.
8. Ju CI, Lee SM. Complications and management of endoscopic spinal surgery. *Neurospine*. 2023;20(1):56-77. <http://doi.org/10.14245/ns.2346226.113>. PMID:37016854.
9. Aygun H, Abdulshafi K. Unilateral biportal endoscopy versus tubular microendoscopy in management of single level degenerative lumbar canal stenosis: a prospective study. *Clin Spine Surg*. 2021;34(6):E323-8. <http://doi.org/10.1097/BSD.0000000000001122>. PMID:33470660.

10. Baoshan X, Haiwei X, Ning L, et al. Comparative study of unilateral double-channel endoscopy and coaxial large-channel endoscopy in the treatment of lumbar spinal stenosis. *Chin Med J*. [Internet]. 2022 [Accessed: 7/8/2025];102(41). Available from: <https://www.scholarmate.com/S/Cv2Omz>
11. Cheng X, Wu Y, Chen B, Tang J. A comparative study of unilateral biportal endoscopic decompression and percutaneous transforaminal endoscopic decompression for geriatric patients with lumbar lateral recess stenosis. *J Pain Res*. 2023;16:2241-9. <http://doi.org/10.2147/JPR.S413502>. PMID:37409162.
12. Eun DC, Lee YH, Park JO, et al. A comparative analysis of bi-portal endoscopic spine surgery and unilateral laminotomy for bilateral decompression in multilevel lumbar stenosis patients. *J Clin Med*. 2023;12(3):1033. <http://doi.org/10.3390/jcm12031033>. PMID:36769686.
13. Han G, Ma L, Cheng Y, et al. Comparison of effectiveness between unilateral biportal endoscopic and uniportal interlaminar endoscopic decompression in the treatment of lumbar spinal stenosis. *Chin J Reparative Reconstr Surg*. 2024;38(3):324-30. [In Chinese]. <http://doi.org/10.7507/1002-1892.202312029>. PMID:38500426.
14. Wang S, Xu H, Ni W, Huang Q, Wang X. Unilateral versus bilateral balloon kyphoplasty in treatment of osteoporotic vertebral compression fractures: a randomized controlled trial protocol. *Medicine*. 2020;99(25):e20524. <http://doi.org/10.1097/MD.00000000000020524>. PMID:32569172.
15. He BL, Zhu ZC, Lin LQ, et al. Biportal versus uniportal endoscopic technique in Unilateral Laminectomy for Bilateral Decompression (ULBD) for lumbar spinal stenosis. *Asian J Surg*. 2024;47(1):112-7. <http://doi.org/10.1016/j.asjsur.2023.05.068>. PMID:37331857.
16. Heo DH, Son SK, Eum JH, Park CK. Fully endoscopic lumbar interbody fusion using a percutaneous unilateral biportal endoscopic technique: technical note and preliminary clinical results. *Neurosurg Focus*. 2017;43(2):E8. <http://doi.org/10.3171/2017.5.FOCUS17146>. PMID:28760038.
17. Lee MH, Jang HJ, Moon BJ, et al. Comparative outcomes of biportal endoscopic decompression, conventional subtotal laminectomy, and minimally invasive transforaminal lumbar interbody fusion for lumbar central stenosis. *Neurospine*. 2024;21(4):1178-89. <http://doi.org/10.14245/ns.2448830.415>. PMID:39765263.
18. Min WK, Kim JE, Choi DJ, Park EJ, Heo J. Clinical and radiological outcomes between biportal endoscopic decompression and microscopic decompression in lumbar spinal stenosis. *J Orthop Sci*. 2020;25(3):371-8. <http://doi.org/10.1016/j.jos.2019.05.022>. PMID:31255456.
19. Ouyang JY, Yang QY, Chen LL, et al. A comparative analysis of unilateral biportal endoscopic and open laminectomy in multilevel lumbar stenosis. *Front Neurol*. 2024;15:1409088. <http://doi.org/10.3389/fneur.2024.1409088>. PMID:39777310.
20. Qing P, Guo W, Xie S, et al. Clinical efficacy of one-hole split endoscopy vs. unilateral biportal endoscopy for the treatment of single-segment lumbar spinal stenosis: a retrospective study with 2-year follow-up. *Front Surg*. 2025;12:1495741. <http://doi.org/10.3389/fsurg.2025.1495741>. PMID:40040816.
21. Sun H, Zhang Q, Xu K, et al. A retrospective study on safety and clinical outcomes of unilateral biportal endoscopic technique for spinal degenerative diseases. *PeerJ*. 2025;13:e19076. <http://doi.org/10.7717/peerj.19076>. PMID:40191755.
22. Tan B, Yang QY, Fan B, Xiong C. Decompression via unilateral biportal endoscopy for severe degenerative lumbar spinal stenosis: A comparative study with decompression via open discectomy. *Front Neurol*. 2023;14:1132698. <http://doi.org/10.3389/fneur.2023.1132698>. PMID:36908592.
23. Wei R, Liu W, Yu M, et al. Delta large-channel endoscopy versus unilateral biportal endoscopy decompressive laminectomy for lumbar spinal stenosis: a prospective randomized controlled trial. *J Orthop Surg*. 2025;20(1):10. <http://doi.org/10.1186/s13018-024-05409-0>. PMID:39754230.
24. Zhou S, A J, Xu X, et al. Comparison of surgical invasiveness and hidden blood loss between unilateral double portal endoscopic lumbar disc extraction and percutaneous endoscopic interlaminar discectomy for lumbar spinal stenosis. *J Orthop Surg Res*. 2024;19(1):778. <http://doi.org/10.1186/s13018-024-05274-x>. PMID:39567947.
25. Choi DJ, Kim JE, Jung JT, et al. Biportal endoscopic spine surgery for various foraminal lesions at the lumbosacral lesion. *Asian Spine J*. 2018;12(3):569-73. <http://doi.org/10.4184/asj.2018.12.3.569>. PMID:29879787.
26. Ito Z, Shibayama M, Nakamura S, et al. Clinical comparison of unilateral biportal endoscopic laminectomy versus microendoscopic laminectomy for single-level laminectomy: a single-center, retrospective analysis. *World Neurosurg*. 2021;148:e581-8. <http://doi.org/10.1016/j.wneu.2021.01.031>. PMID:33476779.
27. Kang T, Park SY, Kang CH, Lee SH, Park JH, Suh SW. Is biportal technique/endoscopic spinal surgery satisfactory for lumbar spinal stenosis patients?: A prospective randomized comparative study. *Medicine*. 2019;98(18):e15451. <http://doi.org/10.1097/MD.00000000000015451>. PMID:31045817.
28. Kim SK, Kang SS, Hong YH, Park SW, Lee SC. Clinical comparison of unilateral biportal endoscopic technique versus open microdiscectomy for single-level lumbar discectomy: a multicenter, retrospective analysis. *J Orthop Surg Res*. 2018;13(1):22. <http://doi.org/10.1186/s13018-018-0725-1>. PMID:29386033.
29. Kim HS, Choi SH, Shim DM, Lee IS, Oh YK, Woo YH. Advantages of new endoscopic Unilateral Laminectomy for Bilateral Decompression (ULBD) over conventional microscopic ULBD. *Clin Orthop Surg*. 2020;12(3):330-6. <http://doi.org/10.4055/cios19136>. PMID:32904063.
30. Lee HG, Kang MS, Kim SY, et al. Dural injury in unilateral biportal endoscopic spinal surgery. *Global Spine J*. 2021;11(6):845-51. <http://doi.org/10.1177/2192568220941446>. PMID:32762357.
31. McGrath LB, White-Dzuro GA, Hofstetter CP. Comparison of clinical outcomes following minimally invasive or lumbar endoscopic unilateral laminotomy for bilateral decompression. *J Neurosurg Spine*. 2019;30(4):491-9. <http://doi.org/10.3171/2018.9.SPINE18689>. PMID:30641853.
32. Rieger B, Sitoci-Ficici KH, Reinshagen C, et al. Endoscopic and microscopic segmental decompression via translaminar crossover spinal approach in elderly patients. *World Neurosurg*. 2019;125:e361-71. <http://doi.org/10.1016/j.wneu.2019.01.078>. PMID:30703594.
33. Park MK, Park SA, Son SK, Park WW, Choi SH. Clinical and radiological outcomes of unilateral biportal endoscopic lumbar interbody fusion (ULIF)

compared with conventional posterior lumbar interbody fusion (PLIF): 1-year follow-up. *Neurosurg Rev.* 2019;42(3):753-61. <http://doi.org/10.1007/s10143-019-01114-3>. PMID:31144195.

34. Pao JL, Lin SM, Chen WC, Chang CH. Unilateral biportal endoscopic decompression for degenerative lumbar canal stenosis. *J Spine Surg.* 2020;6(2):438-46. <http://doi.org/10.21037/jss.2020.03.08>. PMID:32656381.

35. Tang Z, Tan J, Shen M, Yang H. Comparative efficacy of unilateral biportal and percutaneous endoscopic techniques in unilateral laminectomy for bilateral decompression (ULBD) for lumbar spinal stenosis. *BMC Musculoskelet Disord.* 2024;25(1):713. <http://doi.org/10.1186/s12891-024-07825-z>. PMID:39237948.

36. Ding H, Han X, Xing Y, Liu Y, He D, Han X. Clinical and radiological comparison of unilateral biportal endoscopic and percutaneous transforaminal endoscopic discectomy in the treatment of lumbar spinal degenerative disease. *Orthop Surg.* 2025;17(4):1105-13. <http://doi.org/10.1111/os.14361>. PMID:39854041.

37. Pao JL. Preliminary clinical and radiological outcomes of the “no-punch” decompression techniques for unilateral biportal endoscopic spine surgery. *Neurospine.* 2024;21(2):732-41. <http://doi.org/10.14245/ns.2448376.188>. PMID:38955542.

38. Ross N, Aleman C, Dhenin A, Vassal M, Lonjon G. Tubular versus unilateral biportal endoscopy: MRI analysis after unilateral laminectomy for bilateral decompression in lumbar spinal stenosis. *Eur Spine J.* 2025;34(7):2972-80. <http://doi.org/10.1007/s00586-025-08953-3>. PMID:40448852.

39. Kalanchiam GP, Kaliya-Perumal AK, Sampath L, Boey ETH, Oh JYL. Complications in uniportal vs unilateral biportal endoscopic decompression for lumbar spinal stenosis: a scoping review. *Global Spine J.* 2025;30:21925682251346413. <http://doi.org/10.1177/21925682251346413>. PMID:40448305.

40. Xie Y, Gu H, Yongcun W, et al. A comparison between accurate unilateral puncture paths planned by preoperative and conventional unilateral puncture techniques in percutaneous vertebroplasty. *Comput Math Methods Med.* 2022;2022:6762530. <http://doi.org/10.1155/2022/6762530>. PMID:35832135.

41. Li YS, Chen CM, Hsu CJ, Yao ZK. Complications of unilateral biportal endoscopic lumbar discectomy: a systematic review. *World Neurosurg.* 2022;168:359-368.e2. <http://doi.org/10.1016/j.wneu.2022.10.038>. PMID:36527215.

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Complications Resulting from the Use of Cervical Collars in the Context of Trauma: an integrative literature review

Complicações Decorrentes do Uso do Colar Cervical no Contexto Traumático: uma revisão integrativa da literatura

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ABSTRACT

Introduction: The cervical collar is a device widely used for immobilizing the cervical spine in trauma victims, aiming to prevent secondary neurological injuries. However, its prolonged or inadequate use can trigger several clinical complications. **Objective:** To analyze the main complications resulting from the use of the cervical collar in trauma patients. **Methods:** The search was performed in the PubMed and Google Scholar databases, using the descriptors “Cervical collar AND Trauma AND Complications”. Original studies published in the last 10 years, in Portuguese and English, that addressed complications associated with the use of the cervical collar were included. After applying the eligibility criteria, eight studies were selected, including seven cross-sectional studies and one randomized clinical trial. **Results:** Four main groups of complications were identified: neurological, dermatological, respiratory and dysphagia. Among the neurological complications, increased pain sensitivity and elevated intracranial pressure stood out. Dermatological complications included pressure ulcers, with a higher prevalence in the elderly and in cases of prolonged use. In the respiratory aspect, reductions in lung volumes, impairment of vital capacity and an increased incidence of pneumonia. Dysphagia was also reported, with an impact on nutrition, hydration and quality of life. **Conclusion:** Despite the importance of the cervical collar in protecting the spine, its judicious use, with continuous monitoring, is essential in order to reduce risks and optimize clinical outcomes for patients.

Keywords: Spinal cord injuries; Cervical collar; Device-related complications

RESUMO

Introdução: O colar cervical é um dispositivo amplamente utilizado na imobilização da coluna cervical em vítimas de trauma, visando a prevenção de lesões neurológicas secundárias. No entanto, seu uso prolongado ou inadequado pode desencadear diversas complicações clínicas. **Objetivo:** Analisar as principais complicações decorrentes do uso do colar cervical em pacientes traumatizados. **Métodos:** A busca foi realizada nas bases PubMed e Google Scholar, utilizando os descritores “Cervical collar AND Trauma AND Complications”. Foram incluídos estudos originais publicados nos últimos 10 anos, nas línguas portuguesa e inglesa, que abordassem complicações associadas ao uso do colar cervical. Após a aplicação dos critérios de elegibilidade, oito estudos foram selecionados, incluindo sete transversais e um ensaio clínico randomizado. **Resultado:** Foram apontados quatro principais grupos de complicações: neurológicas, dermatológicas, respiratórias e disfagia. Entre as neurológicas, destacaram-se o aumento da sensibilidade dolorosa e elevação da pressão intracraniana. As dermatológicas incluíram úlceras por pressão, com maior prevalência em idosos e em casos de uso prolongado. No aspecto respiratório, foram observadas reduções nos volumes pulmonares, comprometimento da capacidade vital e aumento da incidência de pneumonia. A disfagia também foi relatada, com impacto na alimentação, hidratação e qualidade de vida. **Conclusão:** Apesar da importância do colar cervical na proteção da coluna, é fundamental a sua utilização criteriosa, com monitoramento contínuo, a fim de reduzir os riscos e otimizar os desfechos clínicos dos pacientes.

Palavras-Chave: Traumatismos da medula espinal; Colar cervical; Complicações relacionadas a dispositivos

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INTRODUCTION

Spinal cord injury (SCI) represents one of the most prevalent traumas of the Central Nervous System associated with spinal cord injury and bone fractures¹. With a prevalence of 1:1000 people, cervical spinal cord injuries can lead to short, medium and long-term complications, such as respiratory, musculoskeletal and neurological disorders². The cervical collar (CC) is a medical device used to immobilize the cervical spine and reduce the risk of secondary spinal cord injury by keeping the head in an anatomically normal position³. The potential disability resulting from SCI generates significant impacts on overall quality of life and represents an economic burden for the family and society, which can be mitigated by the use of the cervical collar⁴.

The immobilization generated by the use of the collar reduces the risk of additional damage to the spine that can lead to motor and sensory impairment and morbidity in the most severe cases⁵. Given this, collars are used in various contexts in the trauma scenario, from spinal clearance obtained through clinical examination to the rehabilitation of neuromuscular or musculoskeletal diseases, as well as surgical interventions⁶. Immobilization is achieved through a rigid support around the cervical spine with adjustable straps and height adjustments and with interface models, which vary significantly between collars and between collar locations, implying effects on the designs of the collars and the materials used in each device⁷.

Complications arising from the use of rigid CC have been reported by several authors⁸⁻¹⁰. Among the complications, increased cervical sensitivity, respiratory restrictions, venous engorgement and even ulcerations may occur given the long period with the device^{8,9}. Recently, it has been demonstrated that the use of rigid collars can cause jugular compression and is related to increased intracranial pressure (ICP)⁶. Furthermore, in the elderly population, the use of rigid cervical CC for more than 24 hours can cause a reduction in the level of consciousness, pressure ulcers and congestive pulmonary conditions¹¹. In the literature, there is a positive view of the use of the non-rigid or soft CC regarding complications¹². Despite the small number of comparative studies between these two devices, a low incidence of secondary neurological impairment after cervical spine fracture is reported in patients who used the non-rigid collar¹³.

Unnecessary use of cervical CC also contributes to the emergence of complications¹⁴. In a study developed in Iran, approximately 43% of cases did not have the expected result after the use of CC¹⁵.

The indiscriminate use of this device, in addition to increasing the risks to the patient's health, increases costs to the health system¹⁶. Among several factors, the inappropriate use of CC is related to the lack of reliable predictors for the severity and extent of cervical injuries, as well as the adequate practices of the medical team in placing the devices¹⁵. Thus, the objective of the literature review is to address the main complications resulting from the use of CC in the traumatic context.

METHODS

This is an integrative review of the literature on complications resulting from the use of cervical collars. The study presented the following steps: choice and adaptation of the theme, selection of descriptors, definition of the databases used, extraction of articles, summary analysis of the articles, analysis of the data collected, synthesis and interpretation of the data and final writing. The searches were carried out in the electronic databases PubMed and Google scholar. The descriptors used were: "Cervical collar AND Trauma AND Complications". The inclusion criteria were articles that were available in full in the databases in the last 10 years, articles in Portuguese and English, studies with complications resulting from the use of cervical collars. The exclusion criteria were case reports, review articles and book chapters. A summary of data selection is seen in Figure 1.

DISCUSSION

Eight studies (Seven cross-sectional studies and one randomized trial) were included, addressing the main complications resulting from prolonged use of CC. This review will discuss neurological, dermatological, respiratory complications and dysphagia. A summary of the main information from the studies can be seen in Table 1.

Neurological complications

Among the most frequent neurological complications associated with the use of CC, the increase in pain sensitivity stands out^{15,17,18}. Hoseini Kasnavieh et al.¹⁵ carried out a study with 114 patients suffering from car trauma who used CC. These authors described

Table 1. Complications resulting from the use of a cervical collar in the context of trauma.

Reference	Type of Study	Objective	Sample	Necklace type	Types of injuries	Complications
Hosseini Kasnavieh et al. ¹⁵	Cross-sectional study	To investigate the need for cervical collars in patients with neck problems	114 patients	Rigid cervical collar	Direct trauma, car accident with car, car accident with motorcycle	Sensitivity (pain or discomfort), distraction injuries, intoxication
He et al. ¹⁷	Randomized Trial	To determine whether a cervical collar is effective and for how long it should be worn after hybrid cervical surgery (HS) combining anterior cervical discectomy and fusion (ACDF) and cervical disc arthroplasty (CDA)	80 patients	Rigid cervical collar	Skin injuries mainly from pressure	Pressure ulcers. Difficulty swallowing; cough. Marginal mandibular nerve palsy with long-term sensory degradation. Impairment of daily activities and sleep and decreased lung capacity and function
Kissmer et al. ⁴	Cross-sectional study	To determine knowledge, attitudes and practices regarding cervical collars in adult trauma patients	2036 patients	Soft and rigid cervical collar	Does not show injury	Creation of unnatural articulation points in the cervical spine. Local tissue disruption and worsening of anxiety
Randall et al. ¹⁸	Cross-sectional study	To identify outcomes of patients discharged home with a rigid cervical collar after negative imaging studies	45 patients	Rigid cervical collar	Direct injury, Direct injury after ejection from vehicle, Motor vehicle accident	Tenderness (pain), rash or skin irritation, problems due to insomnia, and difficult speaking or swallowing
Karacabey and Sanr ¹⁹	Cross-sectional study	Describe the changes in optic nerve sheath diameter after cervical collar placement and analyze these changes depending on time in the C-collar	50 patients	Rigid cervical collar	Injury from falls, motor vehicle injury	Increase in optic nerve sheath diameter after 5 min of use
Ladny et al. ²⁰	Cross-sectional study	To evaluate the influence of cervical spine immobilization with 5 different types of cervical collars on the diameter of the optic nerve sheath measured noninvasively by ultrasound in healthy volunteers	60 patients	Ambu Perfit ACE/ Philly One -Peace Collar / Necloc Collar / NexSplit Plus/ New NECKLITE moldable neck brace	Does not show injury	Philly neck brace appears to cause the maximum increase in optic nerve sheath diameter. The Necklite neck brace has the lowest change in optic nerve sheath diameter
Nakanishi et al. ¹¹	Cross-sectional study	To evaluate whether older trauma patients who wore cervical collars for >24 hours are at increased risk of developing collar-related complications compared with those who wore cervical collars for 24 hours	1154 patients	Rigid cervical collar	Does not show injury	Pressure ulcers and hospital-acquired pneumonia
Ala et al. ²¹	Cross-sectional study	To investigate the effect of collar removal on lung volumes and dyspnea in patients with Glasgow 15	50 patients	Rigid cervical collar	Does not show injury	Spirometric parameters

that 54.4% of the patients presented painful sensitivity or discomfort after using CC. 17. Randall et al.¹⁸, after evaluating the complications of prolonged use of rigid CC in 45 patients suffering from multiple types of injuries, the authors found a 25% rate of painful sensitivity due to the use of CC.

The CC may influence increased ICP¹⁹. Previous studies have described the effect of rigid collar in patients with traumatic brain injury (TBI) as a contributor to elevated ICP and therefore potentially causing further deterioration of their condition^{19,20,22}.

Measurement of the optic nerve sheath diameter (ONSD) by ultrasound is a technique used to assess ICP, with an increase in ONSD indicating an increase in ICP^{23,24}. However, there is an association in the literature between an increase in ONSD and the use of CC in patients hospitalized for trauma^{19,23}. Karacabey and Sanr¹⁹, carried out a prospective study with 50 patients who suffered TBI and used CC type C immediately after the traumatic event. It was found that with prolonged use of CC there was an increase in DBNO. The same result was also verified in the work developed by Ladny et al.²⁰. The authors carried out a study with 60 volunteers, where the relationship between 5 cervical collars (Ambu Perfit ACE/ Philly One -Peace Collar / Necloc Collar / NexSplit Plus/ New necklite moldable neck brace) and ONSD. As a result, all cervical collars were considered restrictive and also presented with DBNO, with the CC Philly being responsible for a greater increase in ONSD compared to the other cervical collars.

Dermatological complications

Dermatological complications have been described in the literature^{25,26}. Kissmer and Morris⁴ conducted a study comparing practices and attitudes towards CC in adult trauma patients in different departments of a hospital unit in South Africa, determining reported complications of cervical collars, such as local tissue rupture. In addition, prolonged application can cause pressure ulcers with an incidence of 6.8 to 38%. The development of this complication impacts general care, the rehabilitation process, and mainly the patient's quality of life. 18. He et al.¹⁷ developed a prospective, randomized control study with the aim of finding physical and functional results. The authors showed that the use of cervical collars for different periods of time can bring many complications to patients, such as skin lesions, with impairment of daily activities, but also respiratory complications such as reduced lung capacity and functionality. Grenier et al.²⁵, showed that cervical collars can lead to serious complications,

such as pressure ulcers and airway difficulties. This was a pre -experimental biomechanical study based on simulation. Furthermore, in another study, Tescher et al.²⁶ compared and verified the restrictiveness and pressure characteristics of the tissue interface of two standard cervical collars and two adjustable cervical collars. A total of 48 adult volunteers (24 men and 24 women) received four types of cervical immobilizers (Aspen, Aspen Vista, Miami J and Miami J Advanced) in random order and presented some pressure-related skin lesions, usually somewhere in the mandible .

Although both soft and rigid collars limit head and neck movement, soft collars mitigate the complications associated with the use of rigid collars, including patient discomfort and soft tissue injury²⁷. Furthermore, excessive mechanical loading from interface pressure and shear over time has an associated risk of skin breakdown, leading to pressure ulcers. Common sites of skin breakdown include the occiput, chin, shoulders, and clavicle. However, interface pressure alone provides limited insight into ulcer risk and may not be directly related to its ability to restrict cervical movement. Several intrinsic and extrinsic factors may contribute to ulceration. In particular, individual head shape and size may affect collar fit according to Russel et al.⁷.

Respiratory complications and dysphagia

According to the literature, respiratory complications have been frequently reported^{18,21}. According to Randall et al.¹⁸ initially, collars can cause indentation marks and pressure ulcers, and collars worn for more than 24 hours were associated with acquired pneumonia in patients over 64 years of age. Another critical problem with the collar was difficulty breathing through the airway. Unnecessary immobilizers can worsen patients' condition by restricting neck movement, obstructing the airway and respiratory tract, as demonstrated in the study by Hosseini Kasnavieh et al.¹⁵. In contrast, for older patients, in particular, immobilization with a collar is uncomfortable, may predispose to the development of pressure ulcers, and impair respiratory function.

Dysphagia was also a frequent complication in some studies. He et al.¹⁷, in their study reported difficulty swallowing, coughing and respiratory distress in their studied patients after the use of a rigid cervical collar. In the study conducted by Randall et al.¹⁸, it was found that, of the 45 patients recruited for the study, approximately 18% reported difficulty sleeping after using the cervical collar and 16% reported difficulty speaking or swallowing.

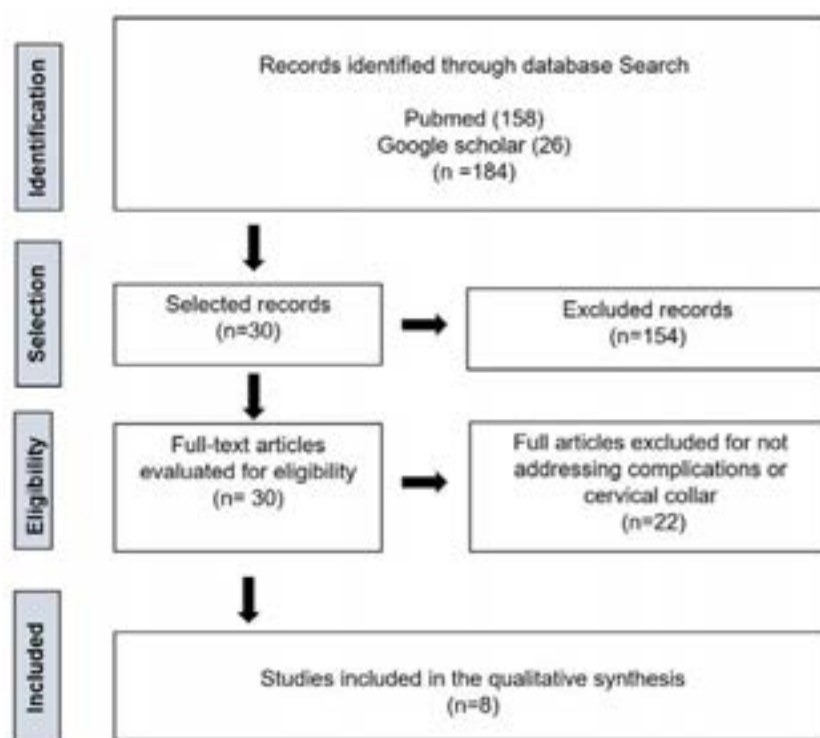


Figure 1. Data selection flow diagram.

The literature also reports changes in spirometric parameters due to the use of this device in the context of trauma. Ala et al.²¹ carried out a study with 50 patients aged between 16 and 65 years, Glasgow 15, victims of trauma, excluding patients with a history of smoking, lung disease, thoracic trauma and multiple trauma. The author found a reduction in Forced Expiratory Volume in the first second (FEV1), mean Forced Expiratory Flow (FEF25–75) and Forced Vital Capacity (FVC). Legg and Cruz²⁸ reported a 3.94% reduction in FVC compared to those who did not use a cervical collar. Ay et al.²⁹, after using the KED model collar, found a reduction in lung function due to dyspnea with reduced saturation observed in some patients.

pain sensitivity and elevated intracranial pressure; dermatological complications, with emphasis on pressure ulcers and skin lesions; respiratory complications, with reduced lung capacity and increased risk of pneumonia, especially in the elderly; in addition to dysphagia and sleep disorders. These findings reinforce the need for careful indication of the device, use for the shortest possible time and strict monitoring of patients, in order to minimize risks and improve clinical outcomes.

CONCLUSION

This review showed that, despite the importance of the cervical collar in preventing secondary injuries in trauma patients, its prolonged or inappropriate use is associated with a wide range of complications. The main complications described in the literature include neurological complications, such as increased

REFERENCES

1. Oliver M, Inaba K, Tang A, et al. The changing epidemiology of spinal trauma: a 13-year review from a Level I trauma centre. *Injury*. 2012;43(8):1296-300. <http://doi.org/10.1016/j.injury.2012.04.021>. PMID:22648015.
2. Behnammoghadam M, Alimohammadi N, Riazi A, Eghbali-Babadi M, Rezvani M. Incidence of cervical collar-related pressure injury in patients with head and neck trauma: A scoping review study. *J Educ Health Promot*. 2023;12(1):252. http://doi.org/10.4103/jehp.jehp_41_23. PMID:37727424.

3. Nutbeam T, Fenwick R, May B, Stassen W, Smith J, Shippen J. Maximum movement and cumulative movement (travel) to inform our understanding of secondary spinal cord injury and its application to collar use in self-extrication. *Scand J Trauma Resusc Emerg Med.* 2022;30(1):4. <http://doi.org/10.1186/s13049-022-00992-9>. PMID:35033151.
4. Kissmer N, Morris D. Knowledge, attitude, and practices regarding cervical collars in adult trauma patients amongst practitioners at three hospitals in KwaZulu-Natal, South Africa. *Afr J Emerg Med.* 2023;13(4):241-4. <http://doi.org/10.1016/j.afjem.2023.09.002>. PMID:37753240.
5. Muzyka L, Bradford JM, Teixeira PG, et al. Trends in prehospital cervical collar utilization in trauma patients: closer, but not there yet. *Acad Emerg Med.* 2024;31(1):36-41. <http://doi.org/10.1111/acem.14822>. PMID:37828864.
6. Joshi H, Hynes LM, Edgell H. Influence of a neck compression collar on cerebrovascular and autonomic function in men and women. *PLoS One.* 2019;14(12):e0225868. <http://doi.org/10.1371/journal.pone.0225868>. PMID:31790493.
7. Russell LJ, Dodd T, Kendall D, et al. A bioengineering investigation of cervical collar design and fit: implications on skin health. *Clin Biomech.* 2024;112:106178. <http://doi.org/10.1016/j.clinbiomech.2024.106178>. PMID:38232471.
8. Ham WH, Schoonhoven L, Schuurmans MJ, Leenen LP. Pressure ulcers, indentation marks and pain from cervical spine immobilization with extrication collars and headblocks: an observational study. *Injury.* 2016;47(9):1924-31. <http://doi.org/10.1016/j.injury.2016.03.032>. PMID:27158006.
9. Núñez-Patiño RA, Rubiano AM, Godoy DA. Impact of cervical collars on intracranial pressure values in traumatic brain injury: a systematic review and meta-analysis of prospective studies. *Neurocrit Care.* 2020;32(2):469-77. <http://doi.org/10.1007/s12028-019-00760-1>. PMID:31190321.
10. Patterson H. Emergency department intubation of trauma patients with undiagnosed cervical spine injury. *Emerg Med J.* 2004;21(3):302-5. <http://doi.org/10.1136/emj.2003.006619>. PMID:15107367.
11. Nakanishi T, Mitra B, Ackland H, O'Reilly G, Cameron P. Time in collars and collar-related complications in older patients. *World Neurosurg.* 2019;129:e478-84. <http://doi.org/10.1016/j.wneu.2019.05.187>. PMID:31150857.
12. Ricciardi L, Stifano V, D'Arrigo S, Polli FM, Olivi A, Sturiale CL. The role of non-rigid cervical collar in pain relief and functional restoration after whiplash injury: a systematic review and a pooled analysis of randomized controlled trials. *Eur Spine J.* 2019;28(8):1821-8. <http://doi.org/10.1007/s00586-019-06035-9>. PMID:31214856.
13. Bäcker HC, Elias P, Braun KF, Johnson MA, Turner P, Cunningham J. Cervical immobilization in trauma patients: soft collars better than rigid collars? A systematic review and meta-analysis. *Eur Spine J.* 2022;31(12):3378-91. <http://doi.org/10.1007/s00586-022-07405-6>. PMID:36181555.
14. Domeier RM, Swor RA, Evans RW, et al. Multicenter prospective validation of prehospital clinical spinal clearance criteria. *J Trauma.* 2002;53(4):744-50. <http://doi.org/10.1097/00005373-200210000-00021>. PMID:12394877.
15. Hoseini Kasnavieh M, Kookli K, Veisi M, Amerzadeh M, Hosseini H, Tahmasebi A. Investigating the rate and affecting factors of unnecessary cervical collar use in trauma patients. *Bull Emerg Trauma.* 2023;11(4):178-83. PMID:38143523.
16. Sundstrøm T, Asbjørnsen H, Habiba S, Sunde GA, Wester K. Prehospital use of cervical collars in trauma patients: a critical review. *J Neurotrauma.* 2014;31(6):531-40. <http://doi.org/10.1089/neu.2013.3094>. PMID:23962031.
17. He J, Liu Q, Yang Z, et al. Cervical collar use following anterior cervical hybrid surgery: protocol for a prospective randomized, time-controlled trial. *Trials.* 2023;24(1):409. <http://doi.org/10.1186/s13063-023-07409-7>. PMID:37328785.
18. Randall MM, Egbert J, Ito BM, Yalung JE, Brown L. Outcomes of patients with negative cervical imaging but persistent neck tenderness discharged with a rigid collar after trauma. *Cureus.* 2022;14(4):e24170. <http://doi.org/10.7759/cureus.24170>. PMID:35592211.
19. Karacabey S, Sanr E. Optic nerve sheath diameter affected by cervical collar placement in minor head trauma patients. *Iran Red Crescent Med J.* 2022;24(10):e188965.
20. Ladny M, Smereka J, Ahuja S, Szarpak L, Ruetzler K, Ladny JR. Effect of 5 different cervical collars on optic nerve sheath diameter: a randomized crossover trial. *Medicine.* 2020;99(16):e19740. <http://doi.org/10.1097/MD.00000000000019740>. PMID:32311968.
21. Ala A, Shams-Vahdati S, Taghizadieh A, et al. Cervical collar effect on pulmonary volumes in patients with trauma. *Eur J Trauma Emerg Surg.* 2016;42(5):657-60. <http://doi.org/10.1007/s00068-015-0565-1>. PMID:26335538.
22. Blaivas M, Theodoro D, Sierzanski PR. Elevated intracranial pressure detected by bedside emergency ultrasonography of the optic nerve sheath. *Acad Emerg Med.* 2003;10(4):376-81. <http://doi.org/10.1197/aemj.10.4.376>. PMID:12670853.
23. Hayreh SS. Optic disc edema in raised intracranial pressure: V. Pathogenesis. *Arch Ophthalmol.* 1977;95(9):1553-65. <http://doi.org/10.1001/archophth.1977.04450090075006>. PMID:71138.
24. Zeiler FA, Unger B, Kramer AH, Kirkpatrick AW, Gillman LM. A unique model for ultrasound assessment of optic nerve sheath diameter. *Can J Neurol Sci.* 2013;40(2):225-9. <http://doi.org/10.1017/S0317167100013779>. PMID:23419572.
25. Grenier G, Despatis MA, Lebel K, Hamel M, Martin C, Boissy P. Removal of the cervical collar from alpine rescue protocols? A biomechanical non-inferiority trial in real-life mountain conditions. *Scand J Trauma Resusc Emerg Med.* 2022;30(1):42. <http://doi.org/10.1186/s13049-022-01031-3>. PMID:35761355.
26. Tescher AN, Rindflesch AB, Youdas JW, et al. Huddleston PM comparison of cervical range-of-motion restriction and craniofacial tissue-interface pressure with 2 adjustable and 2 standard cervical collars. *Spine.* 2016;41(6):E304-12. <http://doi.org/10.1097/BRS.0000000000001252>. PMID:26536441.

27. Wang HN, Campbell J, Doubrovsky A, Singh V, Collins J, Coyer F. Pressure injury development in critically ill patients with a cervical collar in situ: a retrospective longitudinal study. *Int Wound J*. 2020;17(4):944-56. <http://doi.org/10.1111/iwj.13363>. PMID:32239663.

28. Legg SJ, Cruz CO. Effect of single and double strap backpacks on lung function. *Ergonomics*. 2004;47(3):318-23. <http://doi.org/10.1080/0014013032000157878>. PMID:14668165.

29. Ay D, Aktaş C, Yeşilyurt S, Sarıkaya S, Çetin A, Özdoğan ES. Effects of spinal immobilization devices on pulmonary function in healthy volunteer individuals. *Ulus Travma Acil Cerrahi Derg*. 2011;17(2):103-7. <http://doi.org/10.5505/tjtes.2011.53333>. PMID:21644085.

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Clinical Aspects and Therapeutic Management of Cervical Angina: an integrative literature review

Aspectos Clínicos e Manejo Terapêutico da Angina Cervical: uma revisão integrativa de literatura

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ABSTRACT

Introduction: Cervical angina is a rare syndrome characterized by non-cardiac chest pain, often mistaken for angina pectoris. It originates from cervical spine pathologies such as spondylosis and radiculopathy, representing a significant diagnostic challenge in clinical practice.

Objective: To analyze the clinical aspects, complications, and therapeutic approaches of cervical angina through an integrative literature review.

Methods: An integrative review was conducted using the PubMed and CAPES Periódicos databases. Studies published within the last ten years, in Portuguese or English, and available in full text were included. Systematic reviews, book chapters, and review articles were excluded.

Results: Cervical angina presents as anterior chest pain, frequently associated with autonomic symptoms such as dyspnea, vertigo, and headache. The most commonly affected cervical levels are C5-C6 and C6-C7. Diagnosis requires the exclusion of cardiac causes, imaging exams, and specific clinical tests such as Spurling's maneuver. Therapeutic management includes conservative strategies (medication, physiotherapy, chiropractic care) and interventional procedures (injections, nerve blocks), as well as surgical interventions like anterior cervical discectomy and fusion or disc arthroplasty in refractory cases. **Conclusion:** Cervical angina remains an underdiagnosed condition requiring multidisciplinary awareness. Early recognition, combined with individualized therapeutic planning, can reduce patient morbidity and significantly improve quality of life.

Keywords: Cervical angina; Cervicogenic chest pain; Chest pain

RESUMO

Introdução: A angina cervical é uma síndrome rara caracterizada por dor torácica não cardíaca, frequentemente confundida com angina pectoris. Sua origem está associada a patologias cervicais, como espondilose e radiculopatia, representando um desafio diagnóstico relevante na prática clínica. **Objetivo:** Analisar os aspectos clínicos, complicações e condutas terapêuticas da angina cervical por meio de uma revisão integrativa da literatura. **Métodos:** Foi realizada uma revisão integrativa utilizando as bases de dados PubMed e CAPES Periódicos. Foram incluídos estudos publicados nos últimos dez anos, em português ou inglês, disponíveis na íntegra. Excluíram-se revisões sistemáticas, capítulos de livros e artigos de revisão. **Resultados:** A angina cervical manifesta-se como dor torácica anterior, frequentemente acompanhada de sintomas autonômicos como dispneia, vertigem e cefaleia. Os níveis cervicais mais envolvidos são C5-C6 e C6-C7. O diagnóstico exige exclusão de causas cardíacas, exames de imagem e testes clínicos específicos, como a manobra de Spurling. O manejo terapêutico inclui abordagens conservadoras (medicação, fisioterapia, quiropraxia) e intervencionistas (injeções, bloqueios), além de cirurgias como discectomia cervical anterior e artroplastia, indicadas para casos refratários. **Conclusão:** A angina cervical é uma condição subdiagnosticada que demanda atenção multidisciplinar. O reconhecimento precoce, aliado a uma abordagem terapêutica individualizada, pode reduzir morbidades e melhorar significativamente a qualidade de vida dos pacientes.

Palavras-Chave: Angina cervical; Dor torácica cervicogênica; Dor torácica

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INTRODUCTION

Chest pain is an important symptom reported by patients, especially in the emergency context¹. Approximately 30% of patients with acute chest pain actually have cardiological causes, with non-cardiological causes (esophageal, osteoskeletal) diagnosed after excluding cardiac factors². Cervical angina (AC), also known as pseudoangina pectoris, is a condition characterized by chest discomfort and respiratory difficulties similar to cardiac angina resulting from cervical spine pathologies, cervical spondylosis, and radiculopathy³. First described in 1927 by Philips, AC is described as anterior chest pain, retrosternal pain, or epigastric pain exacerbated by exertion and relieved by rest^{4,5}. Sympathetic symptoms such as headache, dizziness, or blurred vision are common⁶. Due to the high frequency of dyspnea and chest pain in various pathologies in patients undergoing emergency care, the diagnosis of angina thoracic becomes a challenge⁷.

There are several mechanisms involved in AC³. Most reported cases are due to compression of the cervical nerve root, particularly C4-8². Lesions at these spinal cord levels affect important nerve structures, which contribute to referred pain and sensory disturbances in the anterior thorax and upper extremities⁸. Furthermore, cortical alterations also influence the presentation of AC^{3,9}. The prefrontal cortex is known to assist in the processing and modulation of complex sensory information related to respiratory and cardiac responses⁹. After sensorimotor alteration due to cervicogenic pathology, the patient may also present angina and dyspnea^{3,9}.

The management of AC becomes a challenge due to its similarity to cardiological alterations¹⁰. Often, the diagnosis is late or overlooked⁵. Both manual and conservative approaches and surgical procedures have been reported^{3,5,11}. Conservative treatment with medication, physical therapy, or immobilization has been shown to be effective¹². Chiropractic treatment, including spinal manipulation and soft tissue mobilization, has led to improvements in pain and quality of life¹³. Regarding the surgical approach to cervical angina, anterior cervical dissection has been reported in the literature to have positive effects also on angioedema symptoms, with better results compared to the conservative approach. Therefore, the objective of this study is to conduct an integrative literature review about cervical angina with focus on clinical aspects, complications, and treatment.

METHODS

This is an integrative literature review on the clinical aspects, complications, and treatment of cervical angina. The study involved the following steps: topic selection and adaptation, keyword selection, definition of the databases used, article extraction, summary analysis, data analysis, data synthesis and interpretation, and final draft. The research was conducted in the PubMed and CAPES Periódicos electronic databases.

The descriptors used were: "Cervical Angina" OR "Cervical Spine-induced Chest Pain" OR "Cervical Radiculopathy" OR "Cervicogenic Chest Pain") AND ("Diagnosis" OR "Etiology" OR "Treatment" OR "Management". The inclusion criteria were: articles that were available in full in the databases in the last 10 years, articles in Portuguese and English, studies with complications resulting from the use of the cervical collar. The exclusion criteria were: systematic review, review articles, book chapters.

DISCUSSION

Clinical and diagnostic aspects

AC represents a rare syndrome with a clinical presentation of chest pain similar to cardiac angina¹⁴. Many of the symptoms can be present both at rest and exacerbated by physical activity¹⁵. The pain is described as sharp or deep, with a squeezing or crushing characteristic, and lasting at least 5 seconds¹⁴. It is usually exacerbated by cervical movement or upper extremity movement¹⁴. Autonomic symptoms, such as dyspnea, dizziness, nausea, pallor, and fatigue, are also frequently seen in patients with cervical angina^{6,14}. Nakajima et al.⁶, conducted a study with 10 patients with suspected cervical angina who were referred for surgical procedures. Of the total, five had retrosternal pain, three had chest pain in the lower left anterior region, and two had epigastric pain. In this study, 50% of the patients included reported symptoms such as dizziness and headache. In the study of Sussman et al.¹⁵, approximately 60% of patients reported autonomic symptoms such as dyspnea, dizziness, diaphoresis, and pallor, which worsened mainly after physical exertion.

Most cases of AC are due to compression of the cervical segment of the spinal cord¹⁵. The most frequently affected levels are

C5-C6 (37%), C6-C7 (30%), C4-C5 (27%), and C3-C4 (4%)¹⁶. The cervical neural roots from C4 to C8 are known to contribute to the sensory and motor innervations associated with anterior chest pain, which may be involved in the primary pathogenesis of cervical angina¹⁷. In addition to compressive causes, spinal ischemia and infarction have also been reported in the literature as causal factors¹⁸. Clinical suspicion should increase whenever the pain is related to neck movements or postures, relieved by a cervical collar or simple head support, and when the patient reports cervicobrachial paresthesias¹. Among the provocative maneuvers, the Spurling maneuver (extension + ipsilateral rotation + axial compression) is the most reproduced: sensitivity of 30–60% and specificity close to 90%, the Jackson maneuver (lateral flexion + compression), the elevated arms test and palpation of trigger points in the scalene, pectoralis major and sternocleidomastoid muscles, important for excluding myofascial pain¹.

Regarding imaging evaluation, anteroposterior, lateral, and oblique radiographs may show loss of lordosis, posterior osteophytes, segmental instability, and narrowing of the disc space; flexion-extension projections reveal occult instability¹⁵. Magnetic resonance imaging is the gold standard: it demonstrates disc herniation, spondylosis, foraminal stenosis, and even intramedullary hyperintense signal on T2-weighted images, an early sign of myelopathy¹⁵. Feng et al.¹, propose five diagnostic criteria: documented exclusion of cardiac disease; reproducible chest pain or pain exacerbated by movement/cervical compression; consistent neurological signs; evidence of cervical pathology on imaging; improvement after spinal-directed treatment (physiotherapy, infiltration, or surgery).

THERAPEUTIC APPROACHES TO CERVICAL ANGINA

From conservative management to surgical innovations

Although the diagnostic challenge of AC is well documented, management decisions are often less clearly defined. In this sense, conservative and interventional cervical angina management strategies stand out. Information regarding therapeutic management is shown in Table 1.

Conservative management: the foundation of treatment

Conservative, non-operative management is universally recognized as the appropriate first-line treatment for AC^{1,15}. The primary goal is to relieve pain, improve function, reduce neuroradicular inflammation, and allow the natural history of the often self-limiting condition to run its course³⁰. A period of conservative treatment of at least three months is recommended before considering surgical intervention, except in cases of severe or progressive neurological deficits^{1,5}.

Pharmacotherapy and immobilization

Pharmacotherapy is used to manage acute symptoms of pain, inflammation, and muscle spasm. Nonsteroidal anti-inflammatory drugs (NSAIDs) reduce inflammation around the compressed nerve root^{18,31}. For patients with significant muscle spasm in the cervical and parascapular regions, muscle relaxants such as cyclobenzaprine may be beneficial²⁵. In cases where the pain is neuropathic, anticonvulsants such as gabapentin, pregabalin, or

Table 1. Main results with conservative and surgical treatment of cervical angina.

Study	Study Type	Sample	Treatment(s)	Key Findings and Outcomes
Kültür and Bal, 2025 ¹⁹	Retrospective Comparative	75 patients	Surgical: CDR vs. ACDF	CDR provided similar symptom relief to ACDF but with better motion and significantly less adjacent segment disease (2.6% vs 16.7%).
Youssef et al., 2025 ²⁰	Retrospective Study	38 patients	Surgical: MI-PCF	Minimally invasive posterior foraminotomy was safe and effective for radiculopathy (94.7% good/excellent outcomes), for both disc and bone issues.
Taso et al., 2025 ²¹	Randomized Controlled Trial	180 patients	Surgical: ACDF. Non-surgical: Structured physical medicine/ rehabilitation and physiotherapy	Surgery (ACDF) is superior for radiculopathy caused by disc herniation but not superior for radiculopathy caused by spondylosis.

Abbreviations: ACDF: Anterior Cervical Discectomy and Fusion. CDA: Cervical Disc Arthroplasty. CDR: Cervical Disc Replacement. CESI: Cervical Epidural Steroid Injection. MDT: Mechanical Diagnosis and Therapy. MI-PCF: Minimally Invasive Posterior Cervical Foraminotomy. PPECFD: Posterior Percutaneous Endoscopic Cervical Foraminotomy and Discectomy. SNRB: Selective Nerve Root Block.

Table 1. Continued...

Study	Study Type	Sample	Treatment(s)	Key Findings and Outcomes
Schultz et al., 2022 ²²	Retrospective Case Series	6.158 patients	Interventional: CESI	Large-scale study (n=12,168) confirmed the safety of epidural steroid injections at all cervical levels, with a very low complication rate (1.1%).
Chu and Wong, 2021 ¹⁰	Case Report	1 patient	Conservative: Chiropractic Care	A patient's angina and radicular symptoms resolved completely with a course of chiropractic treatment (manipulation, soft tissue mobilization, traction).
Chien et al., 2021 ⁵	Retrospective Cohort Study	163 patients	Surgical: ACDF vs. Conservative: Medication, physical therapy, collar immobilization, stellate ganglion block	For refractory cervical angina, ACDF was significantly more effective than conservative care (78.2% vs. 35% good/excellent results).
Shi et al., 2021 ²³	Retrospective Study	26 patients	Diagnostic: SNRB + Surgical: PPECFD	Using a diagnostic nerve block (SNRB) to guide minimally invasive surgery (PPECFD) proved to be a precise and effective strategy (84.6% success).
Al Jammal et al., 2020 ²⁴	Case Report & Systematic Review	1 patient	Surgical: C6-7 CDA	The first reported case of cervical disc arthroplasty for cervical angina resulted in complete and immediate pain resolution.
Htay et al., 2019 ²⁵	Case Report	1 patient	Conservative: Acupuncture, muscle relaxant (tolperisone), and a COX-2 inhibitor (Etoricoxib)	Patient with C6/C7 nerve root compression, a case of cervical angina showed rapid and sustained pain relief with acupuncture, supporting its use in a multimodal conservative approach.
Carlton et al., 2017 ²⁶	Case Report	1 patient	Conservative: MDT (Physical Therapy)	Targeted physical therapy (MDT) was highly effective for radiculopathy, resolving all symptoms even in a patient with a prior failed C5-C7 ACDF surgery.
Kato et al., 2016 ²⁷	Case Report	1 patient	Conservative: Rest, pregabalin, and physical rehabilitation	A case of thoracic disc herniation causing chest pain highlights the need to consider spinal pathology in the differential for non-cardiac chest pain.
Nakajima et al., 2006 ⁶	Retrospective Case Series	10 patients	Surgical: Anterior surgical decompression.	All 10 patients, initially misdiagnosed with heart disease, achieved complete pain relief after surgery, underscoring the diagnostic challenge.
Ozgun and Marshall, 2003 ²⁸	Retrospective Cohort	39 patients	Surgical: ACDF at C6-7	16% of patients with C7 radiculopathy presented with atypical chest or subscapular pain. The vast majority experienced complete or significant symptom relief after surgery.
Jacobs, 1990 ²⁹	Retrospective Case Series	164 patients	Conservative: Hard collar, traction, exercise, medications. Surgical: ACDF for refractory cases (n=38)	The majority of patients responded to conservative treatment, while surgery is highly effective for refractory cases (87% success).

Abbreviations: ACDF: Anterior Cervical Discectomy and Fusion. CDA: Cervical Disc Arthroplasty. CDR: Cervical Disc Replacement. CESI: Cervical Epidural Steroid Injection. MDT: Mechanical Diagnosis and Therapy. MI-PCF: Minimally Invasive Posterior Cervical Foraminotomy. PPECFD: Posterior Percutaneous Endoscopic Cervical Foraminotomy and Discectomy. SNRB: Selective Nerve Root Block.

certain classes of antidepressants, such as tricyclics and SNRIs, are often used^{1,31,32}.

Another approach that can be used is immobilization with a cervical collar for short-term use to limit neck mobility and reduce mechanical irritation of the nerve roots^{1,25}. However, prolonged use is not recommended, as it can lead to atrophy of the cervical muscles and stiffness, impairing long-term recovery³³.

Physiotherapy and rehabilitation

Physical therapy is a fundamental pillar of the conservative strategy, using a multimodal approach to restore function and mitigate pain^{24,33}. Indeed, exercises designed to strengthen the deep cervical flexors and other supporting muscles of the neck and shoulder girdle, as well as stretching exercises to improve flexibility and range of motion, help in the application of specific techniques adapted to the individual clinical presentation of the patient³⁰. Carlton et al.²⁶, describes a 42-year-old woman with persistent neck pain and radiculopathy even after undergoing a surgical procedure at the C5-C7 level. Her symptoms had not resolved with traditional physical therapy, consisting of stretching, strengthening, and traction. However, when she underwent an evaluation using Mechanical Diagnosis and Therapy (MDT), her symptoms were classified as a thoracic spine derangement. A highly specific home exercise program focused on thoracic extension led to the rapid and complete resolution of her symptoms and a full return to function.

Complementary modalities can also play a significant role in a multimodal conservative strategy³⁰. Htay et al.²⁵ details the successful management of a 53-year-old woman with cervical angina caused by C6-C7 nerve root compression. After initial treatments with multiple medications failed to provide relief, she was treated with a combination of acupuncture and targeted medications (a muscle relaxant and a COX-2 inhibitor). The patient experienced significant pain relief within one hour of acupuncture treatment, which was maintained for two months. Chu², in a case report, presents a 56-year-old man with cervicogenic angina and radiculopathy at the C7 level, secondary to cervical spondylosis, who underwent a 3-month multimodal chiropractic treatment, including cervical manipulation, soft tissue mobilization, and intermittent traction, which resulted in complete resolution of thoracic, cervical, and radicular pain symptoms, with an 11-month follow-up radiograph showing increased space of the involved neuroforamen.

In another study by Kato et al.²⁷, a 30-year-old male baseball player with chest wall pain secondary to a T9-T10 thoracic disc

herniation was treated conservatively through an initial rest regimen, administration of pregabalin (150 mg twice daily), and subsequent physical rehabilitation, resulting in a return to full pitching activities 6 months after onset, with a follow-up MRI confirming a reduction in the size of the herniated disc.

Interventional pain management: the bridge between conservative and surgical care

When initial conservative measures fail, minimally invasive interventional procedures serve as an important intermediate step.

Epidural Steroid Injections (ESIs): Cervical ESIs, typically transforaminal, involve the precise administration of a corticosteroid into the epidural space adjacent to the inflamed nerve root. The potent anti-inflammatory effect of the steroid can reduce swelling and nerve irritation, thereby relieving the radicular pain that drives cervical angina³⁴. A retrospective analysis by Schultz et al.²², over a 14-year period of 12,168 fluoroscopy-guided cervical interlaminar epidural steroid injections performed in 6,158 patients at a single pain clinic, revealed a minor complication rate of 1.0% and a major complication rate of 0.06%, with no cases of paralysis or death, demonstrating that complication rates did not increase at spinal levels above C7-T1 and supporting the safety of the procedure in the stepped management of AC.

The retrospective cohort study by Chien et al.⁵, validates its place in the clinical armamentarium and showed that temporary relief of angina was due to blockade of sympathetic afferent nerve transmission.

Surgical treatment: the definitive approach for refractory cases

For patients with disabling symptoms refractory to conservative treatment or with progressive neurological deficits, surgery is the definitive treatment²⁴. The fundamental goal of surgery is to physically decompress the spinal cord and/or nerve roots that are being compressed by the underlying cause, thus eliminating the source of radicular and referred pain¹⁵.

Anterior Cervical Discectomy and Fusion (ACDF): the gold standard

The procedure is performed through an anterior incision, the degenerated disc and osteophytes are removed, and the space is filled with a graft or cage and stabilized with a plate to promote bone fusion³⁵. By directly removing the anterior compressive structures, the procedure addresses the cause of nerve irritation. In

a retrospective cohort study by Chien et al.⁵, comparing 23 patients who underwent ACDF with 140 who received conservative treatment for cervical angina, the surgical group demonstrated superior results. At a two-year follow-up, 78.2% of ACDF patients reported good or excellent results, compared with only 35% in the non-surgical group. This study confirms that for patients with refractory AC, ACDF is a highly effective treatment that provides consistent and long-lasting relief of both angina-like symptoms and associated autonomic dysfunction⁵. Ozgur and Marshall²⁸ report that 16% of 241 patients who underwent anterior cervical discectomy presented with chest pain or subscapularis pain. Of these patients, 90% had significant to complete symptomatic relief after anterior cervical discectomy. Jacobs²⁹ reports 164 patients with cervical angina, most of whom obtained symptomatic relief through conservative treatment. Furthermore, 38 of the 164 (23%) were refractory to conservative treatment and underwent anterior disc excision and spinal fusion. Of these, 33 patients (87%) obtained complete symptomatic relief, and the remaining 5 patients obtained complete relief after subsequent revision surgery²⁹. Nakajima et al.⁶, report 10 surgical patients with cervical pathology who presented with symptoms of pseudoangina. Patients underwent anterior decompression (n = 6) or subtotal spondylectomy (n = 4), and all achieved symptomatic relief within 4 months after the operation.

Cervical Disc Arthroplasty (CDA): the motion preservation alternative

Even though ACDF is highly effective, fusing a spinal segment eliminates motion at that level. This has led to concerns about the potential for accelerated wear, or “adjacent segment disease,” at the levels above and below the fusion. In response to this concern, cervical disc arthroplasty (CDA), or artificial disc replacement, was developed as a motion-preserving alternative. In CDA, after the discectomy is performed, an artificial disc designed to mimic the motion of a natural disc is implanted²⁴. Al Jammal et al.²⁴, presents a 34-year-old man who suffered from persistent neck and chest pain due to a C6-7 disc herniation that was refractory to 12 months of conservative therapy. After a C6-7 ADC, the patient experienced complete and immediate resolution of all his symptoms.

In a retrospective comparative study with a mean follow-up of more than five years of cervical disc arthroplasty (CDA) demonstrated comparable symptomatic relief to Anterior Cervical Discectomy and Fusion (ACDF) regarding pain and disability; however, CDA resulted in significantly better maintenance of segmental range of motion (9.0° versus 1.1°), greater restoration of cervical lordosis

and disc height, and a lower incidence of adjacent segment disease (2.6% versus 16.7%) and revision surgeries¹⁹.

Minimally invasive posterior approaches: foraminotomy and discectomy

Minimally Invasive Posterior Cervical Foraminotomy (MI-PCF) involves making a small incision in the posterior cervical region and removing a small portion of the lamina and facet joint to directly decompress the nerve root. The main advantage of this approach is that it avoids fusion, thus preserving movement without the need for an artificial implant. A retrospective study by Youssef et al.²⁰, evaluated 38 patients who underwent MI-PCF for unilateral radiculopathy and found significant improvements in both pain (Visual Analog Scale) and disability scores (Neck Disability Index), with 94.7% of patients reporting excellent or good results.

In another retrospective study by Shi et al.²³, with a cohort of 26 patients with diagnostically uncertain cervical radiculopathy, treatment with foraminotomy and posterior percutaneous endoscopic cervical discectomy, after identification of the causative nerve root through selective nerve root block, resulted in significant improvements in arm and neck pain scores and the Cervical Disability Index, with 84.6% of patients achieving a good or excellent result by Macnab criteria at a mean follow-up of 14 months and without major complications^{21,23}. Taso et al.²¹, conducted two randomized controlled trials among 180 patients, comparing surgery (ACDF) with a structured, non-surgical program (physical therapy and rehabilitation) for patients with disabling cervical radiculopathy. The crucial distinction was that they stratified patients based on the underlying pathology. In the trial involving patients whose radiculopathy was caused by a cervical disc herniation, surgery resulted in a statistically significant and clinically important improvement in disability (as measured by the Neck Disability Index) compared to non-surgical treatment over 12 months. However, in the second trial, which included patients whose radiculopathy was caused by degenerative spondylosis (bony stenosis), there was no significant difference in outcomes between the surgical and non-surgical groups²¹. Successful management of AC therefore depends on a detailed and patient-specific selection process.

CONCLUSION

AC is a condition that manifests with chest pain similar to cardiac angina, often accompanied by autonomic symptoms

such as dyspnea, headache, and dizziness. Recognition of clinical signs and the use of imaging tests, such as MRI, combined with provocative physical tests, are essential for differential diagnosis, especially in emergency departments where chest pain is highly prevalent and potentially severe. Conservative management with medications, physical therapy, and chiropractic care yields good results in most cases, while surgical approaches, such as anterior cervical discectomy and arthroplasty, are effective in patients refractory to clinical treatment. Cervical angina remains underrecognized, which can delay diagnosis and lead to unnecessary cardiac investigations. Therefore, early identification and appropriate management directly impact patients' quality of life, avoiding additional morbidity, high hospital costs, and misguided treatments.

REFERENCES

- Feng F, Chen X, Shen H. Cervical angina: a literature review on its diagnosis, mechanism, and management. *Asian Spine J.* 2021;15(4):550-6. <http://doi.org/10.31616/asj.2020.0269>. PMID:33108845.
- Chu EC-P. Cervical radiculopathy as a hidden cause of angina: cervicogenic angina. *J Med Cases.* 2022;13(11):545-50. <http://doi.org/10.14740/jmc4025>. PMID:36506762.
- Chu EC-P, Yun S, Huang KHK. Cervicogenic angina and dyspnea secondary to cervical radiculopathy. *Cureus.* 2023;15(4):e37515. <http://doi.org/10.7759/cureus.37515>. PMID:37064724.
- Phillips J. The importance of examination of the spine in the presence of intrathoracic or abdominal pain. *Proc Int Postgrad MA North Am.* 1927;70:3.
- Chien JT, Hsieh MH, Yang CC, Chen IH, Lee RP. Anterior Cervical discectomy and fusion versus conservative treatment for cervical angina conservative treatment. *Clin Spine Surg.* 2021;34(9):E514-21. <http://doi.org/10.1097/BSD.0000000000001178>. PMID:33828047.
- Nakajima H, Uchida K, Kobayashi S, et al. Cervical angina: a seemingly still neglected symptom of cervical spine disorder? *Spinal Cord.* 2006;44(8):509-13. <http://doi.org/10.1038/sj.sc.3101888>. PMID:16331305.
- Lepor NE, McCullough PA. Differential diagnosis and overlap of acute chest discomfort and dyspnea in the emergency department. *Rev Cardiovasc Med.* 2010;11(S2, Suppl 2):S13-23. <http://doi.org/10.3909/ricm11S2S0006>. PMID:20700098.
- Bogduk N, Govind J. Cervicogenic headache: an assessment of the evidence on clinical diagnosis, invasive tests, and treatment. *Lancet Neurol.* 2009;8(10):959-68. [http://doi.org/10.1016/S1474-4422\(09\)70209-1](http://doi.org/10.1016/S1474-4422(09)70209-1). PMID:19747657.
- Higashimoto Y, Sano A, Nishiyama O, et al. Prefrontal cortex activation is associated with dyspnea during methacholine bronchial provocation tests in patients with bronchial asthma. *Allergol Int.* 2020;69(3):453-4. <http://doi.org/10.1016/j.alit.2019.12.005>. PMID:32113986.
- Chu EC-P, Wong AY-L. Cervicogenic dizziness in an 11-year-old girl: a case report. *Adolesc Health Med Ther.* 2021;12:111-6. <http://doi.org/10.2147/AHMT.S341069>. PMID:34866956.
- Bryans R, Descarreaux M, Duranleau M, et al. Evidence-Based guidelines for the chiropractic treatment of adults with headache. *J Manipulative Physiol Ther.* 2011;34(5):274-89. <http://doi.org/10.1016/j.jmpt.2011.04.008>. PMID:21640251.
- Passmore SR, Dunn AS. Positive patient outcome after spinal manipulation in a case of cervical angina. *Man Ther.* 2009;14(6):702-5. <http://doi.org/10.1016/j.math.2009.03.005>. PMID:19380246.
- Thoomes EJ, Scholten-Peters GGM, De Boer AJ, et al. Lack of uniform diagnostic criteria for cervical radiculopathy in conservative intervention studies: a systematic review. *Eur Spine J.* 2012;21(8):1459-70. <http://doi.org/10.1007/s00586-012-2297-9>. PMID:22531897.
- Abe T, Tanei T, Nishimura Y, Saito R. True angina pectoris immediately after cervical disc herniation surgery for preoperative cervical angina symptoms: a case report. *Cureus.* 2022;14(8):e28313. <http://doi.org/10.7759/cureus.28313>. PMID:36168361.
- Sussman WI, Makovitch SA, Merchant SHI, Phadke J. Cervical angina: an overlooked source of noncardiac chest pain. *Neurohospitalist.* 2015;5(1):22-7. <http://doi.org/10.1177/1941874414550558>. PMID:25553225.
- Brodsky AE. Cervical angina: a correlational study with emphasis on the use of coronary arteriography. *Spine.* 1985;10(8):699-709. <http://doi.org/10.1097/00007632-198510000-00003>. PMID:4081876.
- LaBan MM, Meerschaert JR, Taylor RS. Breast pain: a symptom of cervical radiculopathy. *Arch Phys Med Rehabil.* 1979;60(7):315-7. PMID:454130.
- Nakae Y, Johkura K, Kudo Y, Kuroiwa Y. Spinal cord infarction with cervical angina. *J Neurol Sci.* 2013;324(1-2):195-6. <http://doi.org/10.1016/j.jns.2012.11.005>. PMID:23199591.
- Kültür Y, Bal E. Intermediate to long-term clinical and radiological results of cervical disc prosthesis: a comparative study with anterior cervical discectomy and fusion. *Jtss.* 2025;36(2):71-6. <http://doi.org/10.4274/jtss.galenos.2025.59251>.
- Youssef EM, Abdeen M, Saleh MK, Eladawy A, Alaa Eldin AM. Outcomes of minimally invasive posterior cervical foraminotomy for unilateral cervical radiculopathy. *Advanced Spine Journal.* 2025;43(1):3-10. <http://doi.org/10.57055/2974-4822.1314>.
- Taso M, Sommernes JH, Sundseth J, et al. Surgical versus nonsurgical treatment for cervical radiculopathy. *NEJM Evidence.* 2025;4(4):a2400404. <http://doi.org/10.1056/EVIDoa2400404>. PMID:40130970.

22. Schultz DM, Hagedorn JM, Abd-Elseyed A, Stayner S. Safety of interlaminar cervical epidural injections: experience with 12,168 procedures in a single pain clinic. *Pain Physician*. 2022;25(1):49-58. PMID:35051144.
23. Shi C, Xu N, Sun B, et al. Clinical outcomes of posterior percutaneous endoscopic cervical foraminotomy and discectomy assisted with SNRB in treating cervical radiculopathy with diagnostic uncertainty. *Pain Physician*. 2021;24(4):E483-92. PMID:34213874.
24. Al Jammal OM, Diaz-Aguilar LD, Srinivas S, Plonsker J, Sahyouni R, Pham MH. Cervical arthroplasty in the treatment of cervical angina: case report and review of the literature. *Neurospine*. 2020;17(4):929-38. <http://doi.org/10.14245/ns.2040074.037>. PMID:33401872.
25. Htay MNN, Ni H, Moe S. Cervical spondylosis mimicking cardiac angina. *J Case Rep Images Med*. 2019;5: 100050Z09MH2019.
26. Carlton L, Maccio JR, Handford J, Patel P. The application of mechanical diagnosis and therapy in failed anterior cervical discectomy and fusion: a case report. *OROAJ*. 2017;9(4). <http://doi.org/10.19080/OROAJ.2017.09.555766>.
27. Kato K, Yabuki S, Otani K, et al. Unusual chest wall pain caused by thoracic disc herniation in a professional baseball pitcher. *Fukushima J Med Sci*. 2016;62(1):64-7. <http://doi.org/10.5387/fms.2015-25>. PMID:26983590.
28. Ozgur BM, Marshall LF. Atypical presentation of C-7 radiculopathy. *J Neurosurg*. 2003;99(2, Suppl):169-71. <http://doi.org/10.3171/spi.2003.99.2.0169>. PMID:12956459.
29. Jacobs B. Cervical angina. *NY State J Med*. 1990;90(1):8-11. PMID:2296405.
30. Childress MA, Becker BA. Nonoperative management of cervical radiculopathy. *Am Fam Physician*. 2016;93(9):746-54. PMID:27175952.
31. Magnus W, Viswanath O, Viswanathan VK, Mesfin FB. Cervical radiculopathy. Treasure Island: StatPearls Publishing; 2025.
32. Cheshire WP Jr. Spinal cord infarction mimicking angina pectoris. *Mayo Clin Proc*. 2000;75(11):1197-9. <http://doi.org/10.4065/75.11.1197>. PMID:11075751.
33. Brown NJ, Shahrestani S, Lien BV, et al. Spinal pathologies and management strategies associated with cervical angina (pseudoangina): a systematic review. *J Neurosurg Spine*. 2021;34(3):506-13. <http://doi.org/10.3171/2020.7.SPINE20866>. PMID:33276331.
34. Dydyk AM, Hu Y, Stretanski MF, Sekhri NK. Cervical epidural injection. Treasure Island: StatPearls Publishing; 2025.
35. Laratta JL, Reddy HP, Bratcher KR, McGraw KE, Carreon LY, Owens RK 2nd. Outcomes and revision rates following multilevel anterior cervical discectomy and fusion. *J Spine Surg*. 2018;4(3):496-500. <http://doi.org/10.21037/jss.2018.06.16>. PMID:30547110.

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Gliomatosis Cerebri: clinical, radiological, and molecular perspectives on a rare diffuse glioma pattern and report of three cases

Gliomatose Cerebral: perspectivas clínicas, radiológicas e moleculares de um raro padrão de glioma difuso e relato de três casos

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ABSTRACT

Gliomatosis cerebri (GC) is a rare, diffusely infiltrative glial tumor pattern currently defined by the 2021 World Health Organization (WHO) classification as a diffuse glioma involving at least three cerebral lobes, often extending into deep gray matter and infratentorial regions. Clinical presentation varies depending on lesion location. Magnetic resonance imaging (MRI) typically shows widespread T2-FLAIR hyperintensities without significant contrast enhancement or mass effect. A definitive diagnosis requires histopathological confirmation and molecular profiling. Key molecular alterations—including IDH1/2 mutations, 1p/19q codeletion, ATRX loss, and MGMT promoter methylation—are essential for accurate classification, prognosis, and treatment planning. When feasible, safe supramaximal resection is recommended. We present three illustrative cases: two IDH-mutant diffuse gliomas and one IDH-wild-type glioma, each exemplifying the diagnostic complexity and therapeutic challenges associated with GC. These cases underscore the importance of integrating molecular diagnostics within a multidisciplinary framework to optimize clinical decision-making. Furthermore, we highlight the growing relevance of incorporating genomic and metabolomic data into glioma classification to enhance diagnostic accuracy and support personalized therapeutic strategies. This case series encourages the analysis of GC from a genetic and molecular perspective in light of current scientific advances.

Keywords: Gliomatosis cerebri; Diffuse glioma; IDH1 mutation; MGMT methylation; Neuro-oncology; Case report

RESUMO

Gliomatose cerebral (GC) é um padrão tumoral glial raro, difusamente infiltrativo, atualmente definido pela classificação de 2021 da Organização Mundial da Saúde (OMS) como um glioma difuso que envolve pelo menos três lobos cerebrais, frequentemente se estendendo para a substância cinzenta profunda e regiões infratentoriais. A apresentação clínica varia dependendo da localização da lesão. A ressonância magnética (RM) tipicamente mostra hiperintensidades T2-FLAIR generalizadas sem realce significativo pelo contraste ou efeito de massa. Um diagnóstico definitivo requer confirmação histopatológica e perfil molecular. Alterações moleculares importantes — incluindo mutações IDH1/2, codeleção 1p/19q, perda de ATRX e metilação do promotor MGMT — são essenciais para classificação, prognóstico e planejamento terapêutico precisos. Quando viável, recomenda-se a ressecção supramáxima segura. Apresentamos três casos ilustrativos: dois gliomas difusos com mutação IDH e um glioma IDH tipo selvagem, cada um exemplificando a complexidade diagnóstica e os desafios terapêuticos

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associados à GC. Esses casos ressaltam a importância da integração do diagnóstico molecular em uma estrutura multidisciplinar para otimizar a tomada de decisões clínicas. Além disso, destacamos a crescente relevância da incorporação de dados genômicos e metabolômicos na classificação de gliomas para aumentar a precisão diagnóstica e subsidiar estratégias terapêuticas personalizadas. Incentivamos a análise do CG sob uma perspectiva genética e molecular, à luz dos avanços científicos atuais.

Palavras-Chave: Gliomatose cerebral; Glioma difuso; Mutação IDH1; Metilação MGMT; Neuro-oncologia; Relato de caso

INTRODUCTION

Gliomatosis cerebri (GC) is a rare neuro-oncological condition that has undergone substantial redefinition in both its classification and conceptual framework over recent decades¹. Following the revisions introduced in the 2016 and 2021 editions of the World Health Organization (WHO) Classification of Tumors of the Central Nervous System (CNS), GC is no longer recognized as an independent pathological entity. Instead, it is now classified as a diffuse and extensive infiltration pattern^{2,3}. This infiltrative phenotype encompasses both astrocytic and oligodendroglial lineages and typically involves at least three cerebral lobes. In many cases, the process is bilateral and may extend to infratentorial structures, including the brainstem and cerebellum^{2,3}.

A defining feature of GC is the widespread dissemination of neoplastic glial cells throughout the brain parenchyma while preserving the overall cytoarchitectural integrity, thereby differentiating it from the more destructive and necrotic patterns observed in high-grade gliomas^{3,4}. Although no longer classified as a distinct tumor entity by the WHO, the diffuse growth pattern of gliomatosis cerebri presents major diagnostic and treatment challenges, as its widespread brain involvement often hinders extensive surgery and limits the effectiveness of focal radiotherapy⁴.

The true incidence of GC remains poorly characterized, owing to its reclassification and the relative rarity of reported cases. According to data from the Central Brain Tumor Registry of the United States (CBTRUS)⁵, the estimated incidence is approximately 0.15 cases per million individuals. GC has been described across a wide age range (1 to 98 years), with a higher incidence (0.43 per million) reported among older adults (≥ 65 years). A modest male predominance has also been observed, with a male-to-female ratio of approximately 1.4⁴⁻⁶. In Brazil, epidemiological data are notably limited, and national-level registries are lacking,

posing additional barriers to comprehensive understanding¹⁻⁴⁻⁶. As a result, current evidence remains insufficient to establish definitive correlations between specific molecular alterations and this infiltrative phenotype, particularly in the Brazilian context, where data scarcity and reporting inconsistencies persist¹⁻⁶.

CLINICAL CASE PRESENTATIONS

Case 1

A 49-year-old woman presented in January 2023 with her first episode of generalized tonic-clonic seizure. A contrast-enhanced computed tomography (CT) scan of the brain revealed a nonspecific hypodense area in the left parietal lobe. Subsequent magnetic resonance imaging (MRI) demonstrated infiltrative lesions with irregular contours and poorly defined margins, involving both white and gray matter of the superior and inferior left parietal lobes, including the parietal operculum, angular gyrus, supramarginal gyrus, and the left thalamus. These regions exhibited hyperintensity on T2-weighted sequences (Figure 1).

A stereotactic biopsy confirmed the diagnosis of diffuse astrocytoma, IDH1-mutant, (WHO, 2021) (Table 1)³. At presentation, the patient exhibited no neurological deficits: Karnofsky Performance Status – (KPS): 100; Neurologic Assessment in Neuro-Oncology – (NANO): 0. She underwent six cycles of temozolomide (TMZ) in combination with 30 sessions of radiotherapy. Levetiracetam was prescribed at a dose of 1500 mg/day, resulting in effective seizure control.

Fifteen months after the initial diagnosis, the patient experienced seizure recurrence and radiological evidence of disease progression on follow-up MRI, prompting initiation of a new chemotherapy cycle. By 21 months, she exhibited marked clinical deterioration (KPS < 40; NANO: 18). In accordance with her previously

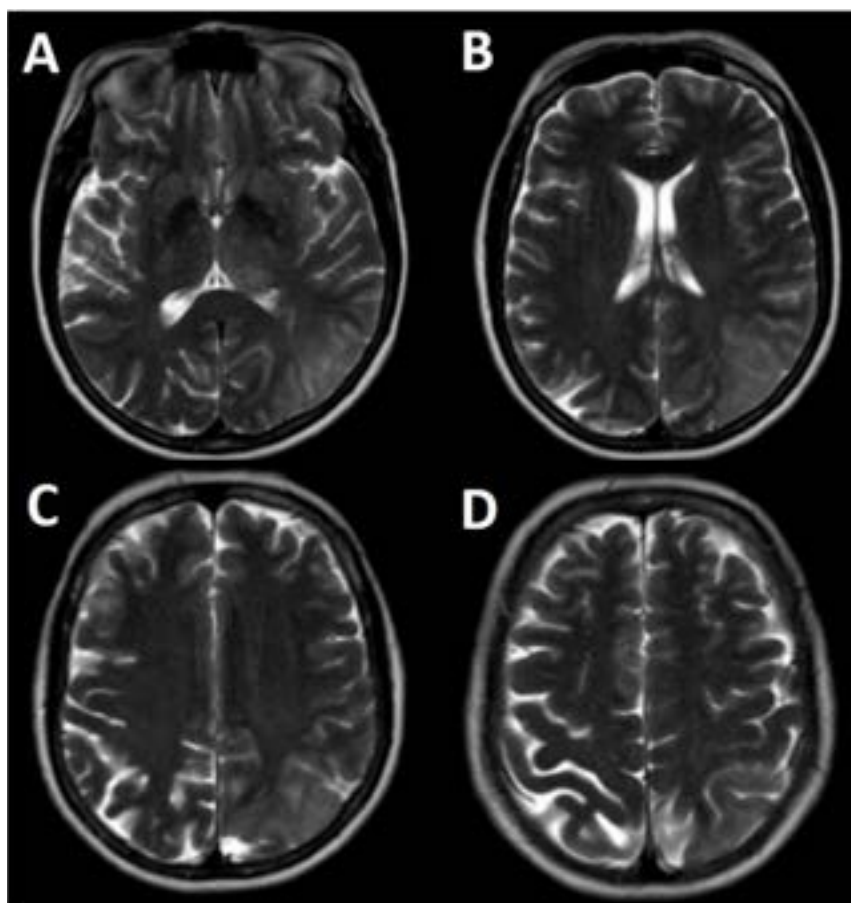


Figure 1. Axial T2-weighted magnetic resonance imaging (MRI) showing a diffuse, hyperintense, and infiltrative lesion involving both cortical (gray matter) and subcortical (white matter) structures of the superior and inferior parietal lobes in the left cerebral hemisphere. The lesion appears ill-defined, with no evident mass effect or significant contrast enhancement, consistent with the radiological features of gliomatosis cerebri.

Table 1. Immunohistochemical profile of three cases of Gliomatosis Cerebri.

Case	Diagnosis	Ki-67 (MIB-1)	GFAP	OLIG2	IDH1	ATRX	TP53
1	Diffuse astrocytoma, IDH-mutant (WHO, 2021)	Positive (<1%)	Positive	Positive (rare cells)	Positive	Inconclusive	Weakly positive (non-aberrant TP53 pattern)
2	High-grade diffuse glioma, IDH-wildtype (Grade 4, WHO, 2021)	Positive (20–25%)	Focally positive	Positive	Negative	Preserved	Negative; Loss of PMS2/MLH1; preserved MSH2/MSH6
3	Astrocytoma, IDH-mutant, Grade 2 (WHO, 2021)	Positive (1%)	Positive	Positive	Positive	Loss (with internal control +)	Strongly positive (diffuse); Aberrant TP53 pattern

Note: Ki-67 is expressed as the percentage of positive tumor cells. ATRX loss is defined by the absence of nuclear staining with preserved internal controls. TP53 is evaluated semi-quantitatively; an aberrant pattern indicates strong diffuse nuclear positivity. Diagnoses are based on the WHO Classification of Tumours of the Central Nervous System (2021)³.

expressed wishes and following a discussion with her family, care was transitioned to a palliative approach.

Case 2

A 64-year-old male patient was admitted presenting with headache, seizures, and motor weakness. Upon examination, he exhibited significant functional impairment, KPS: 40 and NANO score: 14. MRI revealed isolated areas of T2 and FLAIR hyperintensity within the deep white matter of both cerebral hemispheres, accompanied by solid and cystic lesions located in the right temporo-parietal region, affecting both infratentorial and supratentorial compartments (Figure 2). A maximally safe resection of the largest infiltrative lesion was performed via a classical craniotomy centered on the primary mass. Histopathological evaluation confirmed a diagnosis of high-grade diffuse glioma, IDH1-wildtype (WHO Grade 4, 2021)

(Table 1). The patient commenced adjuvant therapy consisting of temozolomide chemotherapy and radiotherapy. Despite treatment, clinical deterioration occurred within three months, necessitating orotracheal intubation. The patient succumbed to the disease four months after initiation of therapy.

Case 3

A 55-year-old male patient presented with a three-month history of severe headaches and behavioral disturbances, including episodes of verbal aggression. During hospitalization, he experienced a seizure. Initial neurological examination revealed no focal deficits (KPS: 100; NANO scale: 0). MRI demonstrated a large area of signal abnormality characterized by mild mass effect, preservation of the cortico-subcortical interface, confluent T2-weighted and FLAIR hyperintensity, and hypointensity on T1-weighted images (Figure 3). A stereotactic biopsy confirmed the diagnosis IDH1-mutant

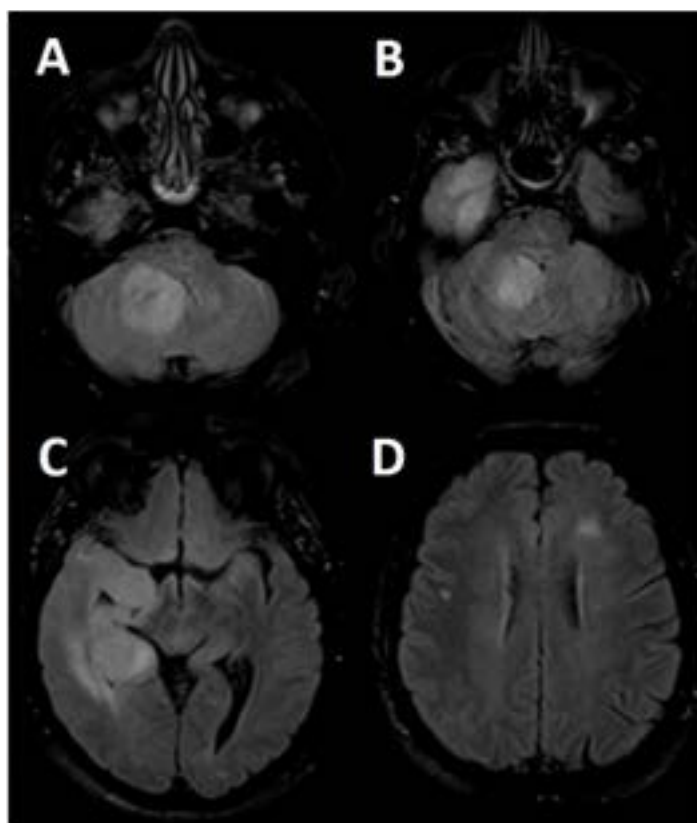


Figure 2. A and B. Axial T2-FLAIR brain MRI showing an expansile and infiltrative lesion centered in the inferomedial aspect of the right cerebellar hemisphere, extending to the superior, middle, and inferior cerebellar peduncles. C and D. Additional infiltrative lesions with similar signal characteristics involve the uncus, temporal pole, inferior temporal gyrus, entire hippocampus, parahippocampal gyrus, medial and lateral occipitotemporal gyri, a portion of the lentiform nucleus, right cerebral peduncle, and a small area of the splenium of the corpus callosum.

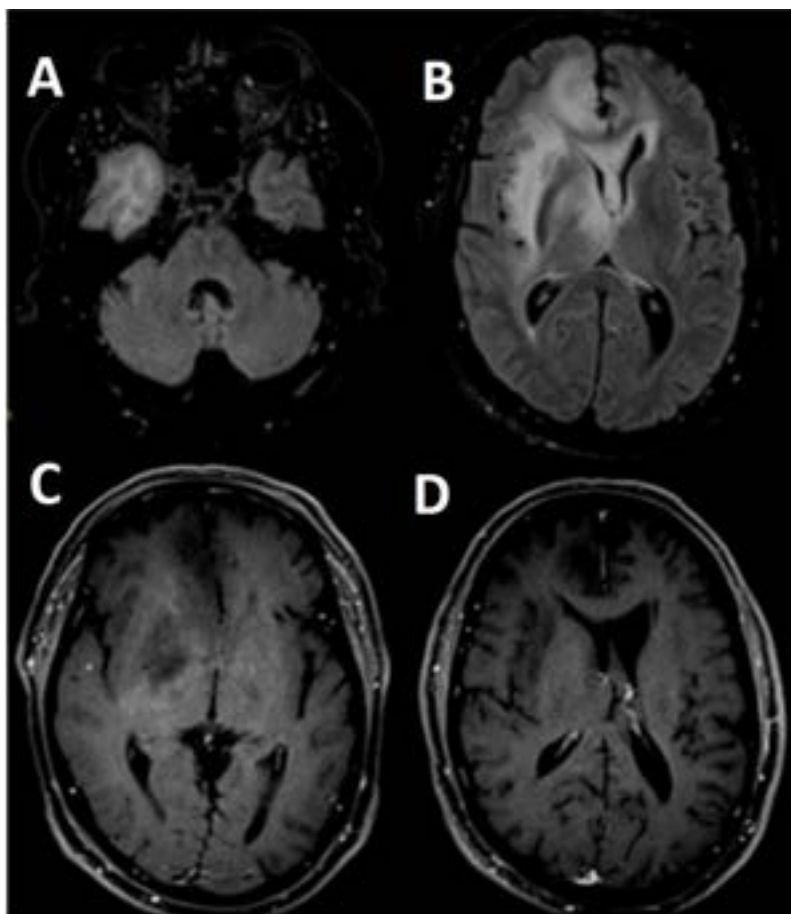


Figure 3. A and B. Magnetic resonance imaging of the brain. Note the confluent hyperintensity on T2-weighted and FLAIR sequences. C and D. And corresponding hypointensity on contrast-enhanced T1-weighted images, centered on the right uncus and striatum. The lesion extends into the anterior limb of the right internal capsule, the base and pole of the right frontal lobe, and crosses the midline via the genu and rostrum of the corpus callosum, reaching the base of the left frontal lobe and the left lentiform nucleus.

astrocytoma, (WHO Grade, 2021), (Table 1). The patient initiated treatment with TMZ followed by radiotherapy. Eight months after diagnosis, he developed clinical symptoms consistent with acute myocardial infarction and rapidly deteriorated into severe heart failure, culminating in death on the same day.

DISCUSSION

Clinical presentation and diagnosis of the gliomatosis cerebri pattern

Under the 2021 WHO classification, GC is no longer recognized as a distinct tumor entity, but rather as a diffuse infiltrative growth

pattern exhibited by certain gliomas¹⁻³. This pattern is defined by the extensive infiltration of neoplastic glial cells involving at least three cerebral lobes, often extending to deep brain structures such as the corpus callosum, basal ganglia, thalamus, brainstem, and cerebellum, typically in a bilateral and asymmetric manner^{1,2}. Despite widespread parenchymal involvement, the overall cytoarchitecture of the brain is frequently preserved¹.

The clinical presentation of GC is markedly heterogeneous and often non-specific, largely reflecting the anatomical distribution and extent of parenchymal infiltration¹. Common manifestations include progressive cognitive decline, memory impairment, personality or behavioral changes, fatigue, and seizures³⁻⁷. Focal neurological deficits, such as hemiparesis, aphasia, visual disturbances, and ataxia, are also frequently observed³.

Involvement of key neuroanatomical pathways, including the corticospinal and spinocerebellar tracts, as well as cranial nerves, may result in myelopathy, papilledema, and cranial neuropathies. Notably, some patients may remain clinically stable or even asymptomatic despite extensive radiological involvement, which can contribute to diagnostic delays or underdiagnosis^{1,7-11}.

Computed tomography (CT) of the head is inadequate for the evaluation of GC; contrast-enhanced MRI remains the gold standard. GC typically presents as diffuse, with no enhancing T2/FLAIR hyperintensities in multiple lobes, often involving the deep gray matter and infratentorial regions¹⁻¹². Features include thickening of the corpus callosum and gray-white matter blurring. Although uncommon, contrast enhancement may suggest anaplastic transformation. Advanced MRI techniques provide additional information about tumor metabolism and extent but cannot definitively distinguish GC from other diffuse pathologies^{12,13}.

Definitive diagnosis of gliomatosis-like infiltration requires histopathological confirmation via stereotactic biopsy. This step is essential for accurately identifying the underlying glioma subtype responsible for diffuse pattern – most commonly IDH-mutant astrocytoma, oligodendroglioma, or glioblastoma. Molecular profiling further enhances diagnostic precision and informs both prognosis and therapeutic strategies¹²⁻¹⁵. Critical biomarkers include IDH mutation, 1p/19q co-deletion, and O⁶-methylguanine-DNA methyltransferase (MGMT) promoter methylation, each of which carries significant prognostic and therapeutic relevance¹⁵.

The prognosis for patients diagnosed with GC remains poor, with reported one-year survival rates ranging from 26 to 52%⁵⁻⁸. Notably, a frequent mismatch exists between clinical symptoms and radiological findings are common; some patients may remain clinically asymptomatic despite imaging evidence of extensive disease burden¹⁻⁶⁻¹⁷. GC can be classified as either primary or secondary. Primary GC is defined involvement of the CNS at initial diagnosis, and it is further subclassified into Type I (classic), characterized by the absence of a discrete mass lesion, and Type II, which presents with a diffuse infiltrative pattern alongside a well-defined tumor mass. Secondary GC refers to the transformation of previously localized glioma and may develop following radiotherapy or antiangiogenic treatment^{6,7-9}.

According to the latest update from the U.S. National Cancer Institute (August 2024), initial management of gliomatosis cerebri

(GC) typically involves maximal safe resection. However, due to the absence of a well-defined mass, most cases are limited to biopsy. No standardized post-biopsy treatment exists; radiotherapy and chemotherapy are commonly used, with decisions tailored to patient-specific factors such as age, tumor burden, histology, and location. Temozolomide remains the most widely used chemotherapeutic agent, particularly given the predominance of astrocytic subtypes in GC¹⁻¹¹.

Molecular profiling in gliomas with a gliomatosis-like growth pattern

Although GC is no longer classified as a distinct pathological entity in the 2021 WHO Classification of CNS tumors, the gliomatosis-like growth pattern – characterized by diffuse and infiltrative involvement of multiple cerebral lobes – continues to hold significant clinical and biological relevance³⁻¹⁸. This growth pattern is most frequently observed in diffusely infiltrating gliomas, including IDH-mutant astrocytomas, oligodendrogliomas, and glioblastomas¹⁸. Consequently, molecular profiling is essential in such cases to enable accurate diagnosis, prognosis assessment, and the development of personalized treatment strategies¹⁹.

Among the most important molecular markers, IDH mutations, particularly in IDH-1 gene, are frequently commonly associated with lower-grade gliomas and secondary glioblastomas, and are correlated with more favorable clinical outcomes. IDH-mutant tumors tend to exhibit slower progression and increased responsiveness to chemotherapy, distinguishing them from their IDH-wildtype counterparts^{3-18,20}. Furthermore, identification of IDH status is critical for differentiating primary from secondary glioblastomas. In recent years, targeted therapies directed against mutant IDH proteins have emerged as promising therapeutic approaches²⁰.

The 1p/19q co-deletion is a key molecular marker for diagnosing oligodendrogliomas and predicts a favorable response to chemotherapy, especially the PCV regimen (procarbazine, lomustine, and vincristine)²¹. It helps differentiate oligodendroglial tumors from astrocytic gliomas, which can also show gliomatosis-like infiltration. MGMT promoter methylation is another critical factor, particularly in glioblastoma patients treated with temozolomide (TMZ)²². This epigenetic modification reduces DNA repair in tumor cells, enhancing the efficacy of alkylating agents and correlating with improved treatment response and survival³⁻²³. Together, these molecular markers are integral to characterizing gliomas with gliomatosis cerebri patterns and guide precision neuro-oncology

approaches consistent with the 2021 WHO molecular classification. Although gliomatosis cerebri lacks a distinct nosological category, these biomarkers remain essential for diagnosis and management of its diffuse, infiltrative glioma subtypes^{3-19,22}.

Clinical implications and prognostic variability: insights from three representative cases

While IDH1 mutations generally indicate a better prognosis in diffusely infiltrating gliomas, outcomes vary due to molecular, anatomical, and treatment factors¹⁹. The cases presented highlight this variability and key diagnostic and management considerations for gliomas with gliomatosis-like growth. In case 1, stereotactic biopsy was performed due to tumor involvement in eloquent brain regions, stressing the need for histopathological confirmation when resection is not possible. Cases 2 and 3 featured open microsurgical resection, allowing wider margins. Maximal safe resection remains the surgical standard when feasible²³.

Despite harboring an IDH1 mutation—typically associated with a more indolent course and favorable response to alkylating agents¹ - the patient in case 1 exhibited clinical and radiological disease progression, with significant functional decline observed 21 months after diagnosis. This case illustrates the biological variability among gliomas with a GC growth pattern and suggests that IDH mutational status alone may be insufficient to predict long-term prognosis.

Case 2 was marked by rapid clinical deterioration, likely influenced by the patient's advanced age at diagnosis, higher histological grade per WHO classification, and baseline clinical impairment, as reflected in the KPS and NANO scales. These factors likely contributed synergistically to the unfavorable outcome³⁻²⁴.

Initial treatment with temozolomide and radiotherapy followed standard protocols for diffusely infiltrating gliomas, with MGMT methylation status guiding therapeutic decisions²². In Case 1, preserved functional status despite eloquent tumor location underscored the value of a patient-centered approach. Case 2 showed rapid deterioration, while Case 3 died of myocardial infarction, emphasizing the need for multidisciplinary care. These outcomes highlight the importance of addressing comorbidities and incorporating other medical specialties into the comprehensive management of glioma patients.

The diffuse and infiltrative nature of gliomas with GC-like growth, especially those involving eloquent or deep brain structures, continues

to limit therapeutic options and negatively impact prognosis¹. A multimodal approach that incorporates molecular profiling, genomics, and advanced neuroimaging may result in better patient outcomes.

CONCLUSION

Following the 2021 revision of the WHO classification of central nervous system tumors, GC is no longer designated as a distinct diagnostic entity and is now recognized as a diffuse growth pattern that can occur in multiple glioma subtypes. Consequently, GC has received less emphasis in recent literature. However, advances in molecular and genomic profiling highlight the need to revisit this highly infiltrative phenotype.

This series presents diffuse gliomas with both IDH1-mutant and wild-type tumors exhibiting a gliomatosis-like growth pattern, characterized by infiltration of more than three lobes, including noncontiguous and eloquent brain areas. Despite advances in molecular diagnostics improving classification and prognosis, clinical outcomes remain variable, and current treatments often have limited effectiveness. These cases underscore the need for early multidisciplinary intervention and tumor board discussions focused on both oncologic control and the patient's quality of life and psychosocial well-being.

Systematic molecular characterization alongside patient-centered multidisciplinary care is essential. This study's main limitations are its small sample size and restricted access to detailed molecular analyses within the Unified Health System (SUS). Further research into the biology of this aggressive infiltrative phenotype is crucial to develop more effective therapies and improve patient outcomes.

REFERENCES

1. Shin I, Sim Y, Choi SH, et al. Revisiting prognostic factors of gliomatosis cerebri in adult-type diffuse gliomas. *J Neurooncol*. 2024;168(2):239-47. <http://doi.org/10.1007/s11060-024-04656-9>. PMID:38700610.

2. Herrlinger U, Jones DTW, Glas M, et al. Gliomatosis cerebri: no evidence for a separate brain tumor entity. *Acta Neuropathol.* 2015;131(2):309-19. <http://doi.org/10.1007/s00401-015-1495-z>. PMID:26493382.
3. Louis DN, Perry A, Wesseling P, et al. The 2021 WHO classification of tumors of the central nervous system: a summary. *Neuro-oncol.* 2021;23(8):1231-51. <http://doi.org/10.1093/neuonc/noab106>. PMID:34185076.
4. Divé I, Weber KJ, Hartung TI, et al. Gliomatosis cerebri (GC) growth pattern: a single-center analysis of clinical, histological, and molecular characteristics of GC and non-GC glioblastoma. *Neurooncol Adv.* 2023;5(1):vdad131. <http://doi.org/10.1093/oaajnl/vdad131>. PMID:38024242.
5. Georgakis MK, Spinos D, Pourtsidis A, et al. Incidence and survival of gliomatosis cerebri: a population-based cancer registration study. *J Neurooncol.* 2018;138(2):341-9. <http://doi.org/10.1007/s11060-018-2802-z>. PMID:29464663.
6. Chen S, Tanaka S, Giannini C, et al. Gliomatosis cerebri: clinical characteristics, management, and outcomes. *J Neurooncol.* 2013;112(2):267-75. <http://doi.org/10.1007/s11060-013-1058-x>. PMID:23341100.
7. Jennings MT, Frenchman M, Shehab T, et al. Gliomatosis cerebri presenting as intractable epilepsy during early childhood. *J Child Neurol.* 1995;10(1):37-45. <http://doi.org/10.1177/088307389501000111>. PMID:7539465.
8. Sanson M, Cartalat-Carel S, Taillibert S, et al. Initial chemotherapy in gliomatosis cerebri. *Neurology.* 2004;63(2):270-5. <http://doi.org/10.1212/01.WNL.0000129985.39973.E4>. PMID:15277619.
9. Taillibert S, Chodkiewicz C, Laigle-Donadey F, Napolitano M, Cartalat-Carel S, Sanson M. Gliomatosis cerebri: a review of 296 cases from the ANOCEF database and the literature. *J Neurooncol.* 2005;76(2):201-5. <http://doi.org/10.1007/s11060-005-5263-0>. PMID:16200347.
10. Mallick S, Giridhar P, Rath GK. Evidence-based practice in neuro-oncology. Cham: Springer; 2022.
11. NATIONAL CANCER INSTITUTE. Gliomatosis cerebri diagnosis and treatment. 2018. Available from: <https://www.cancer.gov>. Accessed: 7/20/2025.
12. Doig D, Thorne L, Rees J, et al. Clinical, imaging and neurogenetic features of patients with gliomatosis cerebri referred to a tertiary neuro-oncology centre. *J Pers Med.* 2023;13(2):222. <http://doi.org/10.3390/jpm13020222>. PMID:36836456.
13. Godoy LFS, Paes VR, Ayres AS, et al. Advances in diffuse glial tumors diagnosis. *Arq Neuropsiquiatr.* 2023;81(12):1134-45. <http://doi.org/10.1055/s-0043-1777729>. PMID:38157879.
14. Beiko J, Suki D, Hess KR, et al. IDH1 mutant malignant astrocytomas are more amenable to surgical resection and have a survival benefit associated with maximal surgical resection. *Neuro-oncol.* 2014;16(1):81-91. <http://doi.org/10.1093/neuonc/not159>. PMID:24305719.
15. Sanson M, Marie Y, Paris S, et al. Isocitrate dehydrogenase 1 codon 132 mutation is an important prognostic biomarker in gliomas. *J Clin Oncol.* 2009;27(25):4150-4. <http://doi.org/10.1200/JCO.2009.21.9832>. PMID:19636000.
16. Lasocki A, Gaillard F. Gliomatosis cerebri: radiological review. *J Med Imaging Radiat Oncol.* 2013;57(6):689-93. <http://doi.org/10.1111/1754-9485.12097>.
17. Takahashi H, Matsutani M. Gliomatosis cerebri: an entity or a pattern? Review of the literature. *J Neurooncol.* 2006;76(3):241-4.
18. Brat DJ, Aldape K, Bridge JA, Canoll P, Colman H, Hameed MR, et al. Molecular Biomarker Testing for the Diagnosis of Diffuse Gliomas. *Arch Pathol Lab Med.* 2022 May 1;146(5):547-574. doi:10.5858/arpa.2021-0295-CP.
19. Aibaidula A, Chan AKY, Shi Z, Li Y, Zhang R, Yang R, et al. Adult IDH wild-type lower-grade gliomas should be further stratified. *Neuro Oncol.* 2017 Oct 1;19(10):1327-1337. doi:10.1093/neuonc/nox078.
20. Mellinghoff IK, Ellingson BM, Touat M, et al. Ivosidenib in IDH1-mutant, chemotherapy-refractory glioma: a phase 1 trial. *Nat Med.* 2023;29(2):250-7.
21. Wesseling P, Capper D. WHO 2021 classification of gliomas. *Neuropathol Appl Neurobiol.* 2022;48(3):e12704. <http://doi.org/10.1111/nan.12704>.
22. Esteller M, Garcia-Foncillas J, Andion E, et al. Inactivation of the DNA-repair gene MGMT and the clinical response of gliomas to alkylating agents. *N Engl J Med.* 2000;343(19):1350-4. <http://doi.org/10.1056/NEJM200011093431901>. PMID:11070098.
23. Bonosi L, Marrone S, Benigno UE, et al. Maximal safe resection in glioblastoma surgery: a systematic review of advanced intraoperative image-guided techniques. *Brain Sci.* 2023;13(2):216. <http://doi.org/10.3390/brainsci13020216>. PMID:36831759.
24. Wu A, Qiu X, An Y, et al. Association of the Neurologic Assessment in Neuro-Oncology (NANO) scale with survival outcomes in patients with gliomas: a systematic review and meta-analysis. *Front Oncol.* 2022;12:836998. <http://doi.org/10.3389/fonc.2022.836998>.

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Endovascular Treatment of Scalp Arteriovenous Fistula: a case report and systematic review of the literature

Tratamento Endovascular das Fístulas Arteriovenosas do Couro Cabeludo: relato de caso e revisão sistemática da literatura

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ABSTRACT

Background: Scalp arteriovenous fistula (AVF), commonly referred to as a cirroid aneurysm, represents an uncommon vascular pathology characterized by abnormal arteriovenous connections within the scalp. Endovascular approach for treatment offers advantages in terms of precision, reduced morbidity, and favorable clinical outcomes when compared to traditional surgical interventions. A systematic review was conducted to examine similar cases, emphasizing diagnostic classification and management strategies. **Case presentation:** A young adult developed a scalp AVF after rubber bullet trauma, confirmed by angiographic imaging. The patient underwent a transarterial endovascular intervention aimed at occluding the fistula. Endovascular treatment led to rapid symptom relief and complete occlusion of the scalp AVF, as verified by follow-up imaging. The systematic review analyzed 42 patients from 14 studies, encompassing all types of scalp arteriovenous fistulae. These cases were characterized by exclusively external carotid artery feeders and scalp vein drainage. All cases were classified according to the Yokouchi classification. A variety of endovascular techniques were employed, including the use of coils, n-butyl cyanoacrylate (NBCA), ethylene vinyl alcohol copolymer (EVOH), and particles, applied via transarterial, transvenous, or direct puncture approaches. In two cases, a second endovascular procedure was required to achieve complete obliteration of the fistula. **Conclusion:** This case highlights the advantages of the endovascular technique in managing scalp arteriovenous fistulas (AVFs), including reduced recovery time and minimized procedural risks. The systematic review demonstrated a variety of endovascular approaches, all achieving complete obliteration of the fistula, further reinforcing the efficacy of this treatment modality for managing scalp AVFs.

Keywords: Arteriovenous fistula; Scalp AVF; Endovascular treatment; Interventional radiology; Cirroid aneurysm

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RESUMO

Introdução: A fistula arteriovenosa (FAV) do couro cabeludo, também conhecida como aneurisma cirsoide, representa uma patologia vascular rara, caracterizada por conexões anômalas entre artérias e veias no couro cabeludo. A abordagem endovascular para o tratamento oferece vantagens em termos de precisão, menor morbidade e desfechos clínicos favoráveis em comparação com as intervenções cirúrgicas tradicionais. Foi realizada uma revisão sistemática da literatura com o objetivo de examinar casos semelhantes, com ênfase na classificação diagnóstica e nas estratégias de manejo. **Relato de Caso:** Um adulto jovem desenvolveu uma FAV do couro cabeludo após trauma por bala de borracha, confirmado por imagem angiográfica. O paciente foi submetido a uma intervenção endovascular transarterial com o objetivo de ocluir a fistula. O tratamento endovascular resultou em alívio sintomático rápido e oclusão completa da FAV, conforme verificado por exames de imagem de seguimento. A revisão sistemática incluiu 42 pacientes de 14 estudos, abrangendo todos os tipos de fistulas arteriovenosas do couro cabeludo. Esses casos apresentavam ramos exclusivamente da artéria carótida externa como fontes de irrigação, com drenagem venosa para veias do couro cabeludo. Todas as fistulas foram classificadas de acordo com a classificação de Yokouchi. Diversas técnicas endovasculares foram utilizadas, incluindo uso de molas, n-butil cianoacrilato (NBCA), copolímero de etileno vinil álcool (EVOH) e partículas, aplicadas por via transarterial, transvenosa ou por punção direta. Em dois casos, foi necessário um segundo procedimento endovascular para obter a oclusão completa da fistula. **Conclusão:** Este caso ressalta as vantagens da técnica endovascular no manejo das fistulas arteriovenosas do couro cabeludo, incluindo menor tempo de recuperação e redução dos riscos do procedimento. A revisão sistemática demonstrou uma variedade de abordagens endovasculares, todas com sucesso na oclusão completa da fistula, reforçando a eficácia dessa modalidade terapêutica para o tratamento das FAVs do couro cabeludo.

Palavras-Chave: *Fístula arteriovenosa; FAV do couro cabeludo; Tratamento endovascular; Radiologia intervencionista; Aneurisma cirsoide*

INTRODUCTION

Scalp arteriovenous fistulas (AVFs) are rare vascular anomalies, typically resulting from trauma. These fistulas form abnormal connections between arteries and veins, often leading to pulsatile vascular lesions and, in certain cases, neurological symptoms¹. While AVFs may be congenital or acquired, traumatic origins, such as penetrating or surgical injuries, are predominant, particularly in regions with dense vasculature like the scalp^{1,2}. Endovascular embolization has been established as an effective treatment for scalp AVFs, allowing for fistula occlusion while preserving normal blood flow². This article presents a case of scalp arteriovenous fistula caused by rubber bullet trauma, successfully treated with endovascular embolization using a balloon catheter and ethylene vinyl alcohol (EVOH).

A systematic review was carried out from November 2024, in PubMed, BVS (Virtual Health Library), and SciELO (Scientific Electronic Library Online), based on PRISMA guidelines to identify similar reports and strict monitoring of inclusion and exclusion criteria, but was not registered in any systematic review database. The search keywords used were: “scalp arteriovenous fistula” OR scalp arteriovenous malformation” OR “cirsoid aneurysm” OR “cirsoid tumor”.

Studies were eligible if they presented case reports or case series reporting on patients with scalp arteriovenous fistula. Studies published from 2015 were selected. We did not adopt any language restrictions to assess the literature comprehensively. Data was extracted by two independent authors and cross-checked by the same authors. Any disagreements were resolved by a third author.

METHODS

Case reports typically do not require Institutional Review Board review as they describe clinical observations without systematic investigation or intent to produce generalizable knowledge. This case includes no identifying information, all data are anonymized, and the patient provided informed consent for publication.

CASE REPORT

A young adult presented with a history of traumatic brain injury (TBI) caused by a rubber projectile approximately two years prior. The trauma resulted in exclusive extracranial injury, which did not require any neurosurgical invasive procedure at the time. Following this period, the patient began to experience

pulsatile vascular dilation in the scalp, particularly in the left temporal region, near to the previous injury site. Upon examination, the patient had a Glasgow Coma Scale (GCS) score of 15, with an atypical gait and visible pulsatile vascular dilation in the scalp.

Digital subtraction angiography (DSA) revealed a large arteriovenous fistula malformation in the scalp, primarily fed by the superficial temporal artery on the left side. There was no involvement of the internal carotid or vertebral arteries. Venous drainage occurred through the superficial veins of the scalp (Figure 1A-B). After multidisciplinary discussion, the decision was made to proceed with endovascular embolization of the lesion.

Under general anesthesia, a femoral puncture was performed using a 6F sheath, and the left external carotid artery was accessed with a 6F guiding catheter. A balloon microcatheter (4×10 mm, (MicroVention, Aliso Viejo, California) was advanced into the posterior branch of the left superficial temporal

artery, positioning it as close as possible to the fistulous point (Figure 1C-D).

With the balloon catheter inflated, the microcatheter was flushed with dimethyl sulfoxide (DMSO), and the fistula was embolized using ethylene vinyl alcohol copolymer (Onyx-18®, Medtronic, Irvine, CA, USA) (Figure 2A-B), performing the “pressure cooker” technique. The embolization successfully resulted in complete occlusion of the arteriovenous fistula while preserving normal blood circulation (Figure 2C). A postoperative fluoroscopy (Figure 2D) and computed tomography (CT) scan revealed no complications, and all liquid embolic agent remained confined to the extracranial compartment.

The patient was admitted to the intensive care unit (ICU) following the embolization procedure, conscious and oriented, with stable vital signs. Routine laboratory tests and CT scan conducted the following day showed no complications related to the procedure. The patient remained stable, with no neurological deficits, and was discharged after a satisfactory evaluation.

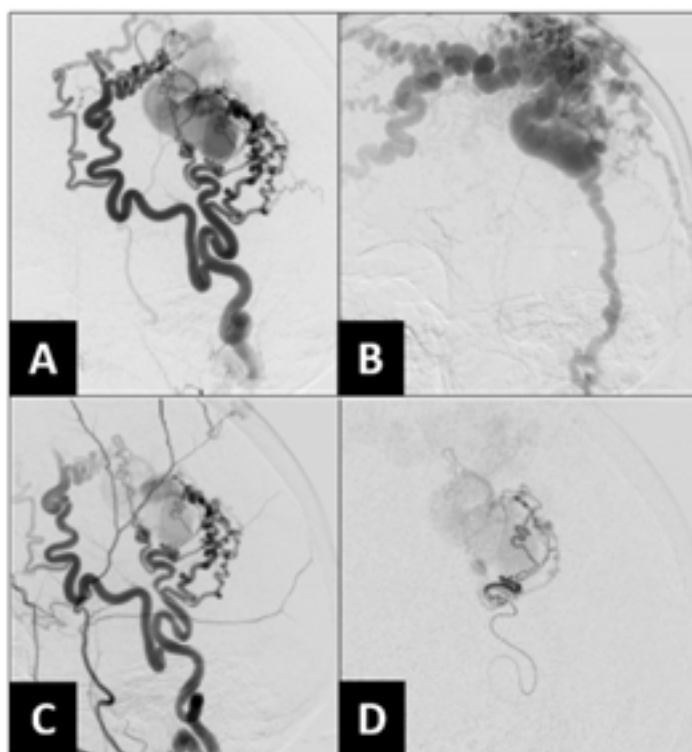


Figure 1. A, B. Digital subtraction angiography (DSA) illustrating an arteriovenous fistula supplied by the left superficial temporal artery, with venous drainage through the superficial scalp veins. C. Endovascular management of the arteriovenous fistula: the posterior branch of the left superficial temporal artery was catheterized using a balloon microcatheter, which was positioned proximally to the fistulous site. D. Microcatheter-based DSA confirming that the catheterized branch was directly supplying the fistulous point.

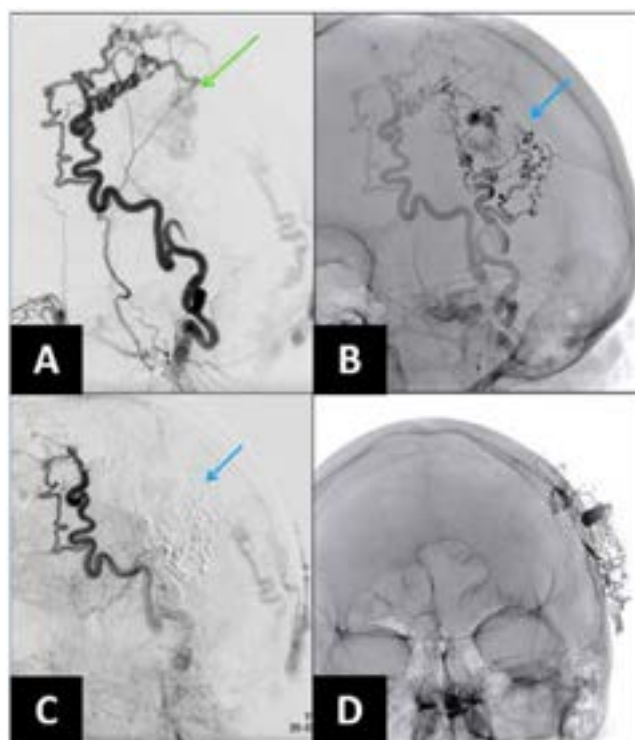


Figure 2. A, B. DSA and fluoroscopic images, respectively, depicting the initial injection of the liquid embolic agent, revealing partial occlusion of the arteriovenous shunt with residual arterial feeders (green arrow) contributing to the fistula. The liquid embolic artifact is indicated by the blue arrow. C. Post-treatment DSA demonstrating complete occlusion of the arteriovenous shunt, with normal circulation preserved and the embolic artifact (blue arrow) confined to the treated area. D. Final fluoroscopy confirming the embolic agent's containment within the extracranial compartment, with no evidence of intracranial artifacts.

RESULTS

The search yielded 93 findings, of which 44 were duplicates and 1 was not available in full text. The remaining articles were screened for publications before 2015 (n=20) and by title and abstract (n=5), resulting in 23 studies eligible for full-text assessment. Finally, fourteen studies were included after excluding 9 papers for presenting intracranial involvement or AVFs not in the scalp (n=7) or no specification of the embolization technique in the case (n=2).³⁻¹⁶ Study selection is depicted in Figure 3.

A total of 42 patient records were eligible. The vessels are described specifically for each case in thirteen^{3-13,15,16} of the fourteen studies³⁻¹⁶, with the feeding arteries and draining veins defining the type of fistula. The following embolization techniques were used in the studies: transvenous (n=5), transarterial (n=5), direct percutaneous puncture (n=30), combined transarterial with transvenous (n=1) and combined transarterial with direct

percutaneous puncture (n=1). In Walker et al.⁷ and Kojima et al.⁸, the “Pressure cooker” technique was used simultaneously, and the “Reinforced concrete” technique was used in Ni et al.¹⁶. In the 14 studies, there was a wide variation in the use of embolic agents, which were individualized in each case. In total, they used Onyx (n=4), n-butyl cyanoacrylate (n=23), SQUID (n=5), PVA (n=1), microcoils (n=1), Penumbra smart coils® (n=1), combined Onyx with balloon catheter (n=1), combined coils with PHIL (n=1), combined Onyx, Scepter C balloon and i-ED 1 ExtraSoft coils (n=1), combined n-butyl cyanoacrylate with coils (n=1) and combined Onyx with coils (n=3). In addition, the type of fistula was represented as type A (n=3), B (n=8) and C (n=6), but there was a loss of data because Gopinath et al.¹⁴ did not specify the type of fistula in each case in the study. The outcomes of the cases correspond to complete resolution of the lesion (n=18), preoperative embolization to reduce intraoperative bleeding (n=2) and the need for surgical excision of the lesion (n=22). Abaunza-Camacho et al.¹⁰ suggested embolization of residual lesions. The present data specified and related to their corresponding cases are summarized in Table 1.

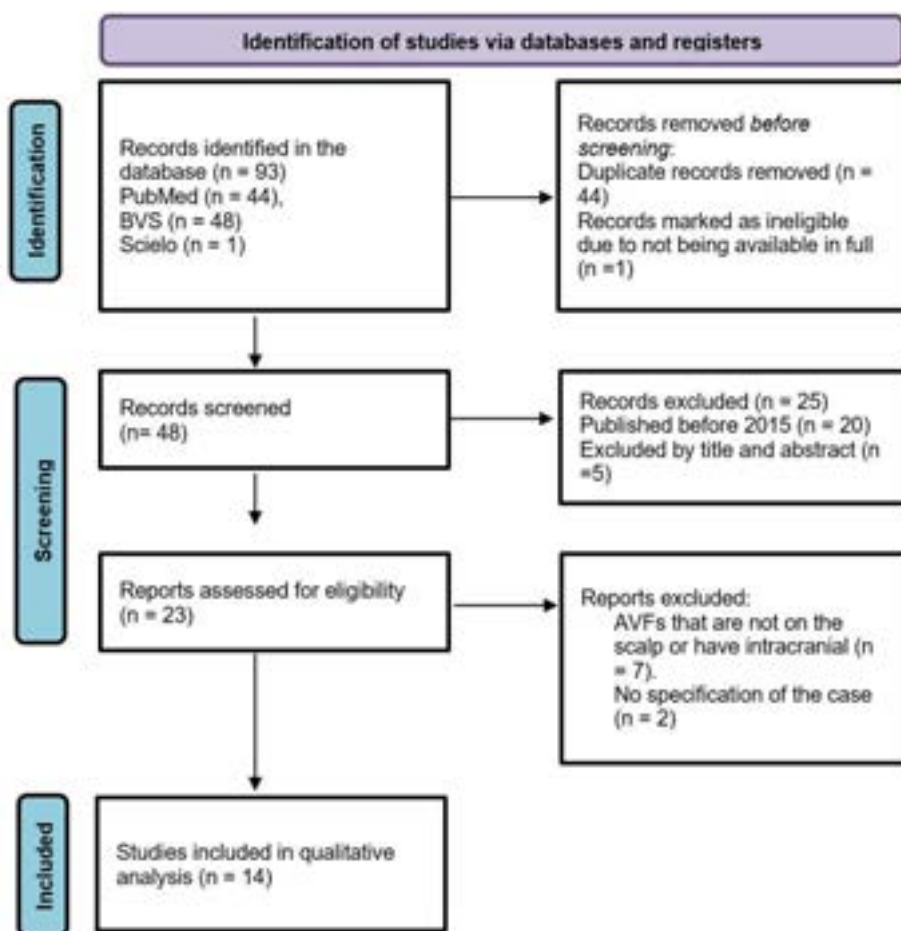


Figure 3. PRISMA flow diagram.

Table 1. Summary of the included studies in Systematic Review.

Author and Year	No. of Patients	AVFs localization	Feeding Arteries	Draining Veins	Technique	Embolitic agent	Conclusion
Kim T et al. ³ , 2024	1	Lt Parietal	Lt; Occipital, middle meningeal, superficial temporal, supraorbital and vertebral	Lt Superficial temporal and internal jugular	Transvenous	Onyx-18 and balloon catheter	Type B Fistula. Transarterial technique failed due to tortuosity of feeders. Transvenous was possible because of a single large draining vein. Onyx was chosen because of its ability to occlude the shunt from the venous outlet, a process facilitated by non-adhesive properties, but must be associated with a balloon catheter to avoid embolism. Lesion completely obliterated and patient did not present recurrence or symptoms at follow-up.

No: number; AVFs: arteriovenous fistulas; AVF: arteriovenous fistula; Lt: left; Rt: right; Onyx: onyx liquid embolic system; PHIL: precipitating hydrophobic injectable liquid; NBCA: n-butyl-2-cyanoacrylate; SQUID: squid liquid embolic system; PVA: Polivinil álcool; STV: superficial temporal vein.

Table 1. Continued...

Author and Year	No. of Patients	AVFs localization	Feeding Arteries	Draining Veins	Technique	Embolic agent	Conclusion
Hsieh YY et al. ⁴ , 2024	1	Rt Temporal	Bilateral superficial temporal	Rt Superficial temporal	Transarterial (Rt side)	NBCA	Type B Fistula. Transarterial technique failed due to contralateral feeding development. NBCA is adhesive and was probably chosen because of the low flux fistula and fast acting properties. Patient suffered post-procedure headache due to venous obliteration. Subsequent surgery was performed, and complete lesion excision was achieved with less intraoperative bleeding. No symptoms, complications or recurrence presented on follow-up.
Subhan M et al. ⁵ , 2023	1	Rt Parietal and extending from the vertex to the right temporal fossa.	Rt External Carotid	Rt Superficial temporal and internal jugular	Transarterial	Microcoils	Type C Fistula. Transarterial technique achieved partial obliteration due to minor collateral development 2 weeks later. Coils were used because of a more controlled release and reduced risk of distal migration, but are less effective in this lesion due to collateral formation. Embolization was used to reduce intraoperative bleeding, but complete obliteration can happen. Patient later had surgery with complete lesion excision and significantly reduced intraoperative blood loss. No symptoms, complications or recurrence presented on follow-up.
Alfaro AJQ et al. ⁶ , 2023	1	Rt Parieto-occipital	Rt Superficial temporal and occipital	Rt Superficial Temporal	Direct percutaneous puncture	Coils and PHIL	Type B Fistula. Direct puncture technique is used in these cases of superficial fistulas with large venous pouches and/or difficult access through other methods, allowing a more localized approach and faster procedure. This was the first described case of the use of PHIL in scalp AVF, its justification includes easy use, cosmetic properties, less glare in imaging and deeper penetration, which may be useful in transvenous approach. Lesion was obliterated and patient presented no symptoms, complications or recurrence on follow-up.

No: number; AVFs: arteriovenous fistulas; AVF: arteriovenous fistula; Lt: left; Rt: right ; Onyx: onyx liquid embolic system; PHIL: precipitating hydrophobic injectable liquid; NBCA: n-butyl-2-cyanoacrylate; SQUID: squid liquid embolic system; PVA: Polivinil álcool; STV: superficial temporal vein.

Table 1. Continued...

Author and Year	No. of Patients	AVFs localization	Feeding Arteries	Draining Veins	Technique	Embolic agent	Conclusion
Walker GB et al. ⁷ , 2023	1	Rt Parieto-temporal	Rt Superficial temporal	Rt Superficial Temporal, Posterior Auricular and eventually Superior Sagittal Sinus	Direct percutaneous puncture with "Manual Pressure Cooker"	SQUID	Type A Fistula. Direct puncture technique was used because of superficial fistula with large venous pouches and difficult access. During embolization, feeder artery was manually compressed, to improve embolic agent penetration, similar to the pressure cooker technique. SQUID, a non-adhesive embolic agent was chosen due to a safe location wherein reflux would not be harmful. The lesion was completely obliterated, and the patient presented no symptoms, complications or recurrence on follow-up.
Kojima D et al. ⁸ , 2022	1	Lt Temporal	Rt Superficial temporal and deep anterior temporal	Rt Superficial temporal and deep temporal	Transarterial with pressure cooker and Transvenous	Onyx, Scepter C balloon and i-ED 1 ExtraSoft coils	Type C Fistula. Transarterial pressure cooker technique achieved partial obliteration because of difficult access through feeders, which needed transvenous approach to complement. In transvenous, coils were used to limit unnecessary Onyx diffusion. However, the Onyx cast diffused to multiple subcutaneous veins and migrated closer to the orbit, needing manual compression of venous outlets which were successful. The lesion was completely obliterated, and the patient presented no symptoms, complications or recurrence on follow-up.
Shi Y et al. ⁹ , 2022	2	Frontal	Bilateral frontal superficial temporal and ophthalmic	Bilateral orbital and facial	Direct percutaneous puncture	Onyx-18	Type B and A Fistula. Both cases opted for direct puncture technique because of difficulty in transarterial approach. Onyx was used because of non-adhesive properties, and manual compression of venous outlets were done to avoid unnecessary diffusion. In both cases the lesion was completely obliterated, and the patient presented no symptoms, complications or recurrence on follow-up.
		Rt Occipital	Rt Ophthalmic	Rt Temporal and Occipital	Direct percutaneous puncture	Onyx-18	

No: number; AVFs: arteriovenous fistulas; AVF: arteriovenous fistula; Lt: left; Rt: right ; Onyx: onyx liquid embolic system; PHIL: precipitating hydrophobic injectable liquid; NBCA: n-butyl-2-cyanoacrylate; SQUID: squid liquid embolic system; PVA: Polivinil álcool; STV: superficial temporal vein.

Table 1. Continued...

Author and Year	No. of Patients	AVFs localization	Feeding Arteries	Draining Veins	Technique	Embolic agent	Conclusion
Abaunza-Camacho JF et al. ¹⁰ , 2021	1	Lt Orbitozygomatic	Lt Ophtalmic, Vidian and external carotid	Lt Ectatic superficial scalp	Direct percutaneous puncture	Onyx and microvascular resection	Type C Fistula. Emergency embolization due to eyelid bleeding related to blunt trauma to a previous fistula. The first embolization achieved 60% of flow reduction, but quickly presented bleeding recurrence, needing a second attempt, which resulted in almost total obliteration. A small residual AVF remained in the eyelid region, feeded by the Vidian and ophtalmic arteries. Patient presented no symptoms, complications, visual sequelae and bleeding recurrence. Follow-up for residual lesion was suggested.
Ordaz JD et al. ¹¹ , 2021	1	Lt Frontotemporal	Deep temporal, superficial temporal and ophtalmic	Scalp veins	Transvenous	Penumbra smart coils®	Type B Fistula. Transvenous technique and use of coils were used because of high flux fistula with large venous pouch, being preferred instead of transarterial technique and/or use of liquid embolic agents, to guarantee a safe embolic positioning and reduce risk of migration.
Furtado SV et al. ¹² , 2021	2	Lt Parieto-occipital	Lt Occipital	Scalp veins	Transarterial	NBCA	Type B fistula. Transarterial technique used due to the predominance of arterial and only venous reflux. Use of transarterial insertion of NBCA reconstituted with lipiodol through the left occipital artery. After embolization, the patient underwent total excision of the lesion.
		Rt Occipital	Rt Occipital	Rt Occipital and Posterior Auricular	Transarterial	SQUID-18	Type B fistula. Transarterial technique used due to the predominance of arterial and only venous reflux. Use of transarterial insertion of Squid through the right occipital artery. There was complete resolution of the fistula after embolization, without the need for surgery.
Sofela A et al. ¹³ , 2020	1	Lt forehead	Lt Superficial temporal	Scalp veins	Transarterial	Onyx	Type A fistula. Transarterial technique used due to the predominance of arterial and only venous reflux. Using transarterial Onyx insertion. There was complete resolution of the fistula after embolization, without the need for surgery.

No: number; AVFs: arteriovenous fistulas; AVF: arteriovenous fistula; Lt: left; Rt: right ; Onyx: onyx liquid embolic system; PHIL: precipitating hydrophobic injectable liquid; NBCA: n-butyl-2-cyanoacrylate; SQUID: squid liquid embolic system; PVA: Polivinil álcool; STV: superficial temporal vein.

Table 1. Continued...

Author and Year	No. of Patients	AVFs localization	Feeding Arteries	Draining Veins	Technique	Embolic agent	Conclusion
Gopinath M et al. ¹⁴ , 2019	25	Left frontal (16%), Right frontal (16%), Left parietal (20%), Right parietal (12%), Left occipital (8%), Right occipital (4%), Midline occipital (4%), Left temporal (12%) and Right temporal (8%).	Did not specify for each case	Did not specify for each case	Direct percutaneous puncture	84% NBCA, 12% SQUID and 4% PVA	The types of fistulae were not specified. The approach was percutaneous by direct puncture due to the presence of a dilated superficial vein and the high efficacy of this technique in high-flow fistulas. This choice simplified the procedure and enabled the direct use of NBCA, SQUID and PVA, which achieved complete occlusion with adequate flow control. The majority of cases required post-embolization surgery (80% of cases), and there is no pattern following surgery, which can be related to incomplete remission of the mass, persistence of symptoms and aesthetic outcome.
Sousa LHA et al. ¹⁵ , 2018	1	Lt Frontal	Superficial temporal, middle meningeal and anterior ethmoidal	Scalp veins	Transarterial and direct percutaneous puncture	NBCA and coils	Type B Fistula. Transarterial technique used due to the predominance of arterial and only venous reflux. To reduce the high flow, we performed a delivery of coils by direct puncture with an improvised cookie-cutter like instrument, followed by injection of NBCA. There was a partial remission of the fistula, requiring surgery for its complete resection, without complications.
Ni W et al. ¹⁶ , 2017	3	Lt Temporal Rt Temporal Rt Temporal	Lt Superficial Temporal and Occipital Occipital, superficial temporal and facial Rt Superficial temporal	Lt Superficial Temporal Superficial temporal Rt Superficial temporal	Transarterial (1st att). Transvenous (2nd att) Transvenous Transarterial (1st att). Transvenous (2nd att)	Onyx (1st). Onyx and Coils (2nd) Onyx-18 and Coils Onyx (1st). Onyx and Coils (2nd)	All cases involved type C AVFs treated with a transvenous "reinforced concrete" technique. This approach was chosen due to multiple arterial feeders, making arterial occlusion unfeasible without venous access. Coils were used to prevent reflux and create a constrictive structure in the venous pouch, while Onyx-18 filled the superior temporal vein and sealed multiple fistulous connections. This method avoided challenges like insufficient penetration of the embolic agent and thrombosis risk. No surgical resection was performed, and embolization successfully reduced the pulsatile lesion without complications at a 3-month follow-up.

No: number; AVFs: arteriovenous fistulas; AVF: arteriovenous fistula; Lt: left; Rt: right; Onyx: onyx liquid embolic system; PHIL: precipitating hydrophobic injectable liquid; NBCA: n-butyl-2-cyanoacrylate; SQUID: squid liquid embolic system; PVA: Polivinil álcool; STV: superficial temporal vein.

DISCUSSION

Scalp arteriovenous fistulas (AVFs) represent rare, high-flow vascular anomalies characterized by direct communications between arterial and venous systems, bypassing the capillary bed. These shunts result in increased venous pressure and flow, predisposing patients to complications such as venous hypertension, cosmetic deformities, and even rupture in larger or long-standing cases¹⁷. Although most AVFs are acquired, commonly following trauma, surgical procedures, or scalp infections, congenital forms have also been reported. Delayed diagnosis is not uncommon, particularly when the inciting trauma is remote or lacks significant initial intracranial involvement, which often leads to late clinical recognition and intervention^{17,18}. Digital subtraction angiography remains the gold standard for diagnosis, as it allows detailed characterization of the fistula's angioarchitecture and informs therapeutic planning. Clinically, patients may present with a wide spectrum of symptoms, ranging from cosmetic concerns (e.g., pulsatile scalp masses or visible vascular dilations) to neurological manifestations depending on fistula location and hemodynamic impact¹⁹.

Scalp AVFs are typically classified according to their angioarchitectural configuration. The classification proposed by Yokouchi et al.²⁰ delineates three types: Type A (a solitary fistula fed by a single artery), Type B (a solitary fistula fed by multiple arteries), and Type C (multiple fistulas with a plexiform network of feeding arteries converging into a dominant draining vein). This framework not only aids diagnostic clarity but also guides treatment selection. Type A lesions are generally well-suited for transarterial embolization, whereas Type B and C lesions, due to their complexity, often necessitate a combination of transarterial, transvenous, or even surgical approaches¹⁶.

Endovascular embolization has emerged as the preferred treatment modality for scalp AVFs, offering a minimally invasive yet highly effective alternative to open surgical excision^{16,20}. Transarterial embolization is typically employed when arterial feeders are well defined and accessible⁵. In cases with tortuous arterial anatomy or complex venous drainage, transvenous routes may be more appropriate³. Direct percutaneous puncture of the lesion is a valuable alternative in select cases, particularly when traditional access routes are unfeasible or when the lesion has multiple feeders. While this technique provides direct access and enhances embolic control, it also carries a higher risk of local complications such as

hematoma and infection^{6,7}. Therefore, careful patient selection and meticulous procedural planning are paramount.

Selection of embolic agents is crucial and depends on the lesion's vascular characteristics. Ethylene vinyl alcohol (EVOH)-based agents—including Onyx-18, SQUID, and PHIL—are commonly employed due to their deep penetration, stable polymerization, and controlled delivery. Onyx-18 remains the most frequently used, with a robust track record of efficacy in high-flow lesions³. SQUID offers improved injectability and reduced reflux, while PHIL features lower radiopacity, less skin discoloration, and enhanced visualization during follow-up imaging—attributes particularly advantageous in superficial AVFs and transvenous applications⁶. N-butyl cyanoacrylate (NBCA), a rapidly polymerizing adhesive, is a cost-effective option suitable for single-feeder or high-flow fistulas, though it demands precise technique to avoid catheter gluing or unintended embolization²¹. Detachable coils are often used adjunctively, particularly in large venous outflows, to provide mechanical occlusion and structural support¹¹.

Advanced embolization techniques such as the “pressure cooker” and “reinforced concrete” approaches have been increasingly employed in challenging cases. The former involves proximal occlusion using coils or balloons to create a pressurized environment that facilitates deeper penetration of the liquid embolic agent while preventing reflux^{7,8,15}. The latter combines coils and liquid agents to enhance lesion stability and reduce the risk of recanalization^{15,16}. These strategies are particularly valuable when navigating complex vascular architectures or minimizing non-target embolization.

Anatomical accessibility of scalp AVFs also allows for innovative adjunctive techniques, such as manual venous compression during transvenous embolization^{8,9}. By temporarily occluding venous egress, this technique promotes retrograde filling of the nidus and enhances embolic agent penetration while reducing the risk of systemic embolization or spread into unintended vascular territories^{7,8}. Such strategies underscore the importance of integrating anatomical knowledge with procedural finesse.

Technical considerations, including the use of balloon microcatheters during EVOH embolization, are essential to optimize outcomes. Despite full balloon inflation, retrograde reflux may occur due to hemodynamic variables or the non-Newtonian behavior of embolic agents like Onyx. Precision in balloon

positioning, complete occlusion, and vigilant monitoring of reflux dynamics are critical to minimizing adverse events and achieving durable occlusion²².

Although endovascular techniques offer high success rates, complete obliteration is not always possible in a single session. Residual shunting may persist, especially in complex lesions. However, even partial embolization significantly reduces intraoperative bleeding and facilitates subsequent surgical resection, if necessary^{17,23,24}. Microsurgical excision remains a definitive treatment in cases refractory to embolization or those with high recurrence risk. Direct identification and cauterization of fistulous connections under magnification enable precise lesion control and minimize the likelihood of recurrence²³. Long-term surgical outcomes are generally favorable, making this approach a valuable option in comprehensive AVF management²⁵.

The present case aligns with previous reports in several key aspects. Similar to other descriptions, it features a post-traumatic scalp AVF manifesting as a pulsatile mass accompanied by an audible bruit—classic hallmarks of this condition^{5,7,26-28}. Notably, the chronicity observed in our patient—who presented with symptoms eight years after the initial trauma—resonates with previous delayed-onset presentations, such as a case in which a scalp AVF appeared five years after an insidious course²⁸ and another where a progressively enlarging mass emerged three years post-injury⁵. Furthermore, like other cases, the AVF in our patient was localized to the superficial temporal and occipital vascular territories and demonstrated arterial supply from branches of the external carotid artery with venous drainage into the external jugular system^{5,7,28}. This constellation of delayed clinical progression and angioarchitecture underscores the importance of long-term vigilance following craniofacial trauma, even in initially asymptomatic patients.

From a therapeutic standpoint, our dual-agent embolization strategy using Onyx and NBCA aligns with contemporary endovascular practice. While some reported cases required multiple sessions to achieve complete occlusion²⁹, our approach achieved immediate and sustained angiographic cure, with excellent clinical and cosmetic results observed at six-month follow-up. Notably, this case involved feeders from both the superficial temporal and occipital arteries—an anatomical variant less frequently described, as most lesions are fed solely by superficial temporal branches²⁷.

Moreover, the use of dual embolic agents, though effective, is not universally reported. Several series have favored a single-agent approach⁷, whereas our strategy underscores the potential advantage of tailoring embolic choice to lesion complexity and vascular architecture.

In conclusion, this case reinforces the efficacy of endovascular techniques in treating scalp AVFs, particularly when individualized to the patient's anatomy and fistula characteristics. The successful use of a dual-agent approach with complete occlusion and no recurrence highlights the importance of flexible, anatomy-driven strategies in managing these rare but impactful lesions.

STRENGTHS AND LIMITATIONS

This case report and systematic review offer several noteworthy strengths. Anchored in a PRISMA-based methodology, the literature review encompasses 42 patients across 14 studies, providing a comprehensive perspective on embolization techniques and agents. The presented case is enriched by high-resolution imaging and a contemporary endovascular approach, the “pressure cooker” technique utilizing EVOH, enhancing its educational value. The discussion thoughtfully situates the case within established classification frameworks, such as the Yokouchi system, and contributes meaningful clinical insights into the endovascular management of scalp arteriovenous fistulas (AVFs).

Nevertheless, certain limitations should be acknowledged. The case adds limited novelty, as similar presentations and techniques have previously been reported. Moreover, a more explicit comparison with existing literature would have further contextualized its relevance. The absence of a meta-analytic synthesis may reduce the scientific weight of the review, and the exclusion of studies published prior to 2015 could restrict the breadth of the analysis, particularly given the rarity of scalp AVFs. Lastly, the six-month follow-up period may not fully capture long-term outcomes. Despite these limitations, the study remains a valuable clinical reference, particularly for guiding interventional strategies in the treatment of complex superficial vascular lesions.

CONCLUSIONS

The management of scalp arteriovenous fistulas necessitates a tailored, multidisciplinary strategy that accounts for the unique anatomical and clinical intricacies of each case. Endovascular approaches, particularly those employing advanced embolization techniques, continue to represent the foundation of treatment, while surgical intervention remains a valuable adjunct in selected scenarios. Meticulous preprocedural planning and precise technical execution are essential to optimize therapeutic outcomes and mitigate the risk of complications.

REFERENCES

1. May JW Jr, Atkinson R, Rosen H. Traumatic arteriovenous fistula of the thumb after blunt trauma: a case report. *J Hand Surg Am*. 1984;9(2):253-5. [http://doi.org/10.1016/S0363-5023\(84\)80154-9](http://doi.org/10.1016/S0363-5023(84)80154-9). PMID:6715837.
2. Lv X, Li Y, Jiang C, Wu Z. Endovascular treatment of brain arteriovenous fistulas. *AJNR Am J Neuroradiol*. 2009;30(4):851-6. <http://doi.org/10.3174/ajnr.A1436>. PMID:19147710.
3. Kim T, Suh SH. Endovascular treatment of scalp arteriovenous fistula: transvenous onyx embolization with balloon occlusion. *Neurointervention*. 2024;19(3):169-73. <http://doi.org/10.5469/neuroint.2024.00374>. PMID:39389780.
4. Hsieh YY, Chen CC, Tu PH, Yang ST, Liu ZH. Management of traumatic scalp arteriovenous fistula: case report and literature review. *Medicine (Baltimore)*. 2024;103(38):e39764. <http://doi.org/10.1097/MD.00000000000039764>. PMID:39312347.
5. Subhan M, Shah S, Patel S, Ramanathan A. Hybrid Endovascular and Surgical Treatment Scalp Arteriovenous Fistula. *Cureus*. 2023;15(11):e49450. <http://doi.org/10.7759/cureus.49450>. PMID:38152828.
6. Alfaro AJQ, Ortíz AFH, Mejía JA, et al. Traumatic scalp arteriovenous fistula post capillary implantation successfully treated using PHIL embolic agent. *Surg Neurol Int*. 2023;14(12):12. http://doi.org/10.25259/SNI_1002_2022. PMID:36751445.
7. Walker GB, Wang AP, Hadwen J, et al. Direct Puncture of the Superficial Temporal Artery in Embolization of a Scalp Arteriovenous Fistula: A Case Report. *Neurointervention*. 2023;18(1):67-71. <http://doi.org/10.5469/neuroint.2022.00465>. PMID:36717084.
8. Kojima D, Akamatsu Y, Fujimoto K, Oikawa K, Kashimura H, Kubo Y, et al. Utility of manual venous compression during transvenous Onyx injection for a scalp arteriovenous fistula: illustrative case. *J Neurosurg*. Case Lessons. 2022;4(18):CASE22317. [doi:http://doi.org/10.3171/CASE22317](http://doi.org/10.3171/CASE22317)
9. Shi Y, Liu P, Liu Y, et al. Case report: Endovascular treatment of two scalp arteriovenous malformation cases via direct percutaneous catheterization: A case series. *Front Neurol*. 2022;13:945961. <http://doi.org/10.3389/fneur.2022.945961>. PMID:35959410.
10. Abaunza-Camacho JF, Vergara-García D, Pérez F, et al. Emergent Hybrid Treatment of a Ruptured Scalp Arteriovenous Fistula with Eyelid involvement: technical Note. *J Neurol Surg A Cent Eur Neurosurg*. 2021;82(5):490-3. <http://doi.org/10.1055/s-0041-1723848>. PMID:33845513.
11. Ordaz JD, Villelli NW, Bohsntedt BN, Ackerman LL. Delayed presentation of a traumatic scalp arteriovenous fistula. *Surg Neurol Int*. 2021;12(238):238. http://doi.org/10.25259/SNI_263_2021. PMID:34221569.
12. Furtado SV, Srinivasa R, Vala K, Mohan D. Contemporary management of scalp cirroid aneurysm: A dual-trained neurosurgeon's perspective. *Clin Neurol Neurosurg*. 2021;201:106437. <http://doi.org/10.1016/j.clineuro.2020.106437>. PMID:33373833.
13. Sofela A, Osunronbi T, Hettige S. Scalp Cirroid Aneurysms: Case Illustration and Systematic Review of Literature. *Neurosurgery*. 2020;86(2):E98-107. <http://doi.org/10.1093/neuros/nyz303>. PMID:31384940.
14. Gopinath M, Malik V, Sarma PS, Rajan JE, Kannath SK. Results of endovascular-first approach for treatment of scalp arteriovenous malformations and the effect of simplified angiographic classification in therapeutic decision-making and outcomes. *World Neurosurg*. 2019;121:e277-86. <http://doi.org/10.1016/j.wneu.2018.09.092>. PMID:30261380.
15. Sousa LHA, Gatto LAM, Demartini Z Jr, Koppe GL. Scalp cirroid aneurysm: an updated systematic literature review and an illustrative case report. *World Neurosurg*. 2018;119:416-27. <http://doi.org/10.1016/j.wneu.2018.08.098>. PMID:30149169.
16. Ni W, Tian Y, Gu Y, Mao Y. Transvenous endovascular treatment for scalp arteriovenous fistulas: results with combined use of onyx and coils. *World Neurosurg*. 2017;107:692-7. <http://doi.org/10.1016/j.wneu.2017.08.056>. PMID:28838879.
17. Davis AJ, Nelson PK. Arteriovenous fistula of the scalp secondary to punch autograft hair transplantation: angioarchitecture, histopathology, and endovascular and surgical therapy. *Plast Reconstr Surg*. 1997;100(1):242-9. <http://doi.org/10.1097/00006534-199707000-00036>. PMID:9207682.
18. Picard L, Bracard S, Moret J, Per A, Giacobbe HL, Roland J. Spontaneous dural arteriovenous fistulas. *Semin Intervent Radiol*. 1987;4(4):219-41. <http://doi.org/10.1055/s-2008-1076012>.
19. Hage ZA, Few JW, Surdell DL, Adel JG, Batjer HH, Bendok BR. Modern endovascular and aesthetic surgery techniques to treat arteriovenous malformations of the scalp: case illustration. *Surg Neurol*. 2008;70(2):198-203, discussion 203. <http://doi.org/10.1016/j.surneu.2007.04.016>. PMID:18291477.

20. Yokouchi T, Iwabuchi S, Tomiyama A, Samejima H, Ogata N, Goto K. Embolization of scalp AVF. *Interv Neuroradiol*. 1999;5(Suppl 1):121-6. <http://doi.org/10.1177/15910199990050S122>. PMID:20670552.

21. Triano MJ, Lara Reyna J, Schupper AJ, Yaeger KA. Embolic agentes and microcatheters for endovascular treatment of cerebral arteriovenous malformations. *World Neurosurg*. 2020;141:383-8. <http://doi.org/10.1016/j.wneu.2020.06.118>. PMID:32592963.

22. Kular S, Tse G, Pahwa B, et al. Micro-balloon-assisted embolization of anterior cranial fossa dural arteriovenous fistula via a trans-ophthalmic approach - a technical report and case series. *Neuroradiology*. 2022;64(6):1269-74. <http://doi.org/10.1007/s00234-022-02929-6>. PMID:35307749.

23. Yakes WF. Endovascular management of high-flow arteriovenous malformations. *Semin Intervent Radiol*. 2004;21(1):49-58. <http://doi.org/10.1055/s-2004-831405>. PMID:21331109.

24. Beathard GA, Arnold P, Jackson J, Litchfield T, PHYSICIAN OPERATORS FORUM OF RMS LIFELINE. Aggressive treatment of early fistula failure. *Kidney Int*. 2003;64(4):1487-94. <http://doi.org/10.1046/j.1523-1755.2003.00210.x>. PMID:12969170.

25. Belli S, Yabanoglu H, Aydogan C, Parlakgumus A, Yildirim S, Haberal M. Surgical interventions for late complications of arteriovenous fistulas. *Int Surg*. 2014;99(4):467-74. <http://doi.org/10.9738/INTSURG-D-14-00012.1>. PMID:25058786.

26. Lee K, Subhan M, Shah S, Ramanathan A. Hybrid endovascular and surgical treatment of a traumatic scalp arteriovenous fistula. *Cureus*. 2023;15(11):e49450. PMID:38152828.

27. Ordaz JD, Villelli NW, Bohsntedt BN, Ackerman LL. Delayed presentation of a traumatic scalp arteriovenous fistula. *Surg Neurol Int*. 2021;12:238. http://doi.org/10.25259/SNI_263_2021. PMID:34221569.

28. Dalyai RTZ, Schirmer CM, Malek AM. Transvenous balloon-protected embolization of a scalp arteriovenous fistula using Onyx liquid embolic. *Acta Neurochir (Wien)*. 2011;153(6):1285-90. <http://doi.org/10.1007/s00701-011-0998-1>. PMID:21479581.

29. Kato Y, Sano H, Sakai N, Satomi J. Transvenous balloon-protected embolization of a scalp arteriovenous fistula using Onyx liquid embolic. *Acta Neurochir (Wien)*. 2011;153(6):1285-90. PMID:21479581.

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Psychosurgery for Severe Self-Injurious Behavior: case report and literature review

Psicocirurgia para Comportamento Autolesivo Severo: relato de caso e revisão da literatura

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ABSTRACT

Self-injurious behavior (SIB) refers to deliberate acts of self-harm and is often associated with psychiatric, neurological, or a combination of both disorders. When resistant to traditional treatments, psychosurgery can be a viable option for severe cases of SIB. This procedure involves surgical interventions aimed to alter brain function to alleviate debilitating mental health symptoms. This article presents the case of a 31-year-old male with chronic non-progressive encephalopathy and severe SIB, who underwent psychosurgery after years of unsuccessful treatment, alongside a literature review.

Keywords: Psychosurgery; Self-injurious behavior; Stereotaxic technique; Internal capsule; Hypothalamus

RESUMO

O comportamento autolesivo (CAL) refere-se a atos deliberados de autoagressão e está frequentemente associado a transtornos psiquiátricos, neurológicos ou a uma combinação de ambos. Quando resistente aos tratamentos tradicionais, a psicocirurgia pode ser uma opção viável para casos graves de CAL. Esse procedimento envolve intervenções cirúrgicas destinadas a alterar a função cerebral para aliviar sintomas graves de saúde mental. Este artigo apresenta o caso de um paciente de 31 anos com encefalopatia crônica não progressiva e CAL severo, que foi submetido a psicocirurgia após anos de tratamentos mal-sucedidos, juntamente com uma revisão da literatura.

Palavras-Chave: Psicocirurgia; Comportamento autodestrutivo; Técnicas estereotáxicas; Cápsula interna; Hipotálamo

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INTRODUCTION

Self-mutilation or self-injurious behavior (SIB) refers to deliberate acts of self-harm that result in injury, often performed without suicidal intent. Severe forms of SIB, such as eye enucleation and castration, are frequently associated with psychosis and intoxication, while stereotypic forms, like head banging and self-biting, are commonly linked to Tourette's syndrome and severe intellectual disabilities¹. In patients with SIB associated with psychiatric and neurological disorders, psychosurgery has emerged as a treatment option aimed to reduce self-harm in cases resistant to traditional pharmacological and psychological therapies².

This case report details a psychosurgical intervention utilizing a bilateral stereotactic technique, targeting the anterior capsule and posteromedial hypothalamus, in a patient with chronic encephalopathy and severe self-injurious behavior after years of treatment resistance. Previous reports have described patients with both SIB and hetero-aggressive behavior, or only the latter. To date, no documented cases of anterior capsulotomy combined with hypothalamotomy have been performed specifically for self-mutilation, making this the first reported case. This study aims to enhance the understanding of psychosurgery's role in managing complex neuropsychiatric conditions.

CASE PRESENTATION

A 31-year-old male patient with chronic non-progressive encephalopathy and a severe psychiatric disorder exhibits episodes of unprovoked aggressiveness characterized by extreme self-harm. He was born prematurely at 8 months, weighing 2,420 grams and measuring 44 cm, with neonatal complications including hypoxia and cardiopulmonary arrest.

From an early age, he experienced feeding difficulties, unable to properly engage his suckling reflex, which required supplemental feeding through a spoon and dropper, ultimately resulting in malnutrition. At 29 days old, after receiving BCG and hepatitis B vaccinations, he experienced a rare allergic reaction accompanied by severe hypoglycemia, resulting in a 20-day hospitalization.

Throughout the years, he began to exhibit neuropsychiatric disorders. He developed an aversion to touch and social interaction, gravitating towards isolation and dark environments. Since childhood, he engaged in self-harming behaviors, including repeatedly banging his head against furniture corners and injuring his right eye and forehead. His self-harm escalated in adulthood, leading to the mutilation of his eyes, resulting in eventual blindness, and the mutilation of his mouth and nose, resulting in the loss of nasal cartilage and skin (Figure 1). Multiple neurological, psychiatric and psychological treatments were attempted but proven to be unsuccessful.

Due to resistance to multiple treatments and severe progressive episodes of self-harm, his neurological team recommended psychosurgery, which was approved by the Regional Medical Council (CRM) and the Federal Medical Council (CFM). The patient underwent bilateral stereotactic anterior capsulotomy and posteromedial hypothalamotomy. Under general anesthesia, burr holes were made in the paramedian frontal regions, and the procedure involved stimulation at frequencies ranging from 25 to 100 Hz, followed by repeated radiofrequency lesioning at



Figure 1. Mutilation of the mouth and nose.

80°C for one minute. Additional lesions were created above and below the primary target, and this process was subsequently repeated bilaterally in the anterior limb of the internal capsule and hypothalamus. The procedure was uneventful.

A CT scan performed on the first postoperative day revealed small lacunar hypodensities in the anterior limbs of the internal capsules and anterior thalami, with partially defined borders. Bilateral frontal pneumocephalus was also noted, exerting no compressive effects on the adjacent brain parenchyma.

After the procedure, the patient exhibited significant improvement. Two months post-surgery, his self-injurious behavior was effectively managed with previously ineffective medications, and he has not attempted self-harm in the past three years. However, he continues to be bottle-fed and never developed the ability to walk or speak, remaining under regular medical follow-up through home care.

DISCUSSION

The foundations of modern psychosurgery were established in the 19th century, driven by the exploration of surgical interventions for neuropsychiatric disorders. In 1888, Gottlieb Burckhardt performed what is considered the first modern psychosurgery, conducting an experimental topectomy^{3,4}. This procedure involved the topical excision of part of the cerebral cortex, in patients with severe psychiatric conditions⁵. Burckhardt carried out a total of six topectomies, sometimes repeating the procedure on the same patient, targeting predominantly the temporal and parietal lobes based on theories of aggressive behavior. In his 1891 report, he described three procedures as successful, two as partial successes, and one resulting in the patient's death. Facing significant disapproval from his peers, Burckhardt ceased his experiments⁴.

Decades later, in 1935, John Fulton and Carlyle Jacobsen extended this research by experimenting with chimpanzees. Their procedure involved the removal of both frontal lobes, which reportedly resulted in the animals becoming emotionally expressionless and losing their typical responses to unmet rewards³. Following their work, Portuguese neurologist Antonio Egas Moniz, who was the first to coin the term "psychosurgery", proposed ablating

the frontal cortex in individuals with psychiatric disorders^{5,6}. He named the procedure "leucotomy," derived from the Greek words *leuco* (white) and *tomos* (cut), reflecting the cutting of the white matter. Moniz's procedure was first carried out by his collaborator, neurosurgeon Almeida Lima, who injected ethanol into the subcortical white matter of the prefrontal cortex via an opening in the skull. Unsatisfied with the results, Moniz later developed a specialized instrument called a leucotome, a retractable wire loop that could be inserted and rotated to cut neural tissue⁷.

In 1936, Moniz published the results of his procedure on 20 patients who had undergone prefrontal leucotomy⁸. Moniz and Lima's approach showed the most favorable outcomes in patients with anxiety, prolonged depression unresponsive to treatment, severe agitation, and persistent suicidal attempts⁹. The procedure gained significant recognition when Moniz was awarded the Nobel Prize for Physiology or Medicine in 1949 for his discovery of leucotomy's therapeutic value in certain mental disorders^{5,10}.

Also in 1936, Freeman and Watts refined Egas Moniz's technique, introducing the lobotome, a metal rod with a sharp tip, and named their procedure lobotomy, from the Greek words *lobos* (portion) and *tomos* (cut). Unlike Moniz's single incision per hemisphere, their method involved three or more incisions based on the case's severity⁷. The procedure often led to neuropsychiatric complications, such as seizures and "frontal lobe syndrome," characterized by apathy, lack of focus, and socially inappropriate actions⁵. In 1942, Freeman and Watts published a monograph analyzing 200 cases, reporting postoperative improvements in 63% of patients, no change in 23%, and negative outcomes in 14%³.

In 1946, Freeman developed the transorbital lobotomy, eliminating the need for neurosurgeons by using an ice-pick-like orbitoclast to reach the prefrontal cortex through the eye socket, with patients being rendered unconscious through electroshock therapy. Freeman continued to perform the procedure and promoted it through mainstream media, despite complications arising from the use of rudimentary instruments, lack of sterile techniques, inadequate anesthesia, and insufficient perioperative monitoring, as well as fatalities due to intracranial hemorrhage⁵.

In the following years, the practice of psychosurgery became widespread across Western countries. However, throughout the 1950s, it faced growing criticism within the medical community and subsequently from society as a whole due to concerns

about the overlooked risks associated with the procedure⁵. The introduction of psychotropic drugs significantly reduced reliance on psychosurgery due to their improved safety profile and milder adverse effects. By the 1970s, the practice had declined substantially. In the present day, psychosurgery is reserved for severe mental disorders that do not respond to medication and has been enhanced by innovations such as surgical stereotaxy and deep brain stimulation (DBS), as well as more sophisticated techniques and neuroimaging technologies¹¹.

Stereotactic surgery involves the use of a cartesian coordinate system to precisely target areas of the brain for surgical intervention, allowing for the destruction or modulation of small regions with increased precision¹². Deep brain stimulation (DBS), on the other hand, involves the implantation of electrodes in specific brain regions to deliver electrical impulses, usually at high frequencies, modulating abnormal activity. While DBS has shown positive outcomes in treating refractory neurological and psychiatric disorders and offers the advantage of reversibility, its higher cost compared to stereotactic ablation can limit its accessibility¹³.

In Brazil, individuals with mental disorders are protected under Law No. 10,216 and psychosurgical procedures are regulated by The Federal Council of Medicine through Resolution No. 2,057. The recommendation for psychosurgery must be made by the attending physician and validated by a psychiatrist and a neurosurgeon from separate institutions, detailing the diagnosis, treatment duration, and resistance to prior therapies. Furthermore, criteria such as a minimum illness duration of five years and documented treatment resistance must be met, with all procedures recorded in the patient's medical file while ensuring confidentiality^{14,15}.

In this case, a stereotactic technique was employed to bilaterally target the anterior limb of the internal capsule and the posteromedial hypothalamus. The anterior capsulotomy was initially developed by a French neurosurgeon, Jean Talairach in 1949, and later refined by Lars Leksell in 1952⁵. This technique targets the fronto-limbic fibers that traverse the anterior limb of the internal capsule as they pass between the caudate and putamen nuclei of the basal ganglia⁴. Short-term adverse effects, such as headache, confusion, and incontinence, have been reported, while long-term effects, although rare, may include weight gain, fatigue, memory loss, incontinence, and seizures¹⁶. In the present literature, the procedure is more associated with refractory OCD,

being considered an effective treatment option, with a positive side-effect profile¹⁷.

Posteromedial hypothalamotomy, first described in the 1960s, targets the hypothalamus's role in processing external stimuli and coordinating aggressive behavior¹⁸. In animal studies, stimulation of the posteromedial nucleus triggers stereotyped aggression, including claw exposure, piloerection, pupil dilation, and biting, independent of cortical influence¹⁹. The procedure has demonstrated long-term effectiveness in treating patients with refractory aggression, with follow-up studies showing significant improvements over 10-25 years²⁰.

Previous case reports have documented the successful combination of bilateral anterior capsulotomy with hypothalamotomy in treating both SIB and hetero-aggressive behavior, or solely the latter (Table 1)^{11,18,21}. These case reports were derived from an integrative review utilizing the following databases: American Psychological Association, the UCB Magazines Portal, and the Brazilian Journal of Neurosurgery Website (JBNC). Inclusion criteria consisted of articles in Portuguese and English that discussed the combination of bilateral anterior capsulotomy with hypothalamotomy for treating self-injurious behavior and hetero-aggressive behavior, or solely the latter, and that were available in full text. Exclusion criteria included incomplete articles and those unrelated to the study's focus or not meeting the inclusion criteria. This report is the first in which the procedure was performed specifically and only for self-mutilation.

Other procedures including amygdalotomy, anterior cingulotomy, and subcaudate tractotomy, have also been studied for their effectiveness in treating refractory aggression²². However, there are no established parameters to determine the optimal surgical technique and target for refractory SIB. Instead, the decision is based on multiple factors, including equipment availability and patient-specific considerations¹¹.

In this case report, the defining factor leading to the decision for psychosurgery was the patient's escalating history of severe self-mutilation, resistant to conventional treatments, with repeated episodes of life-threatening injuries. Multiple consultations with neurologists, psychiatrists, and neurosurgeons were conducted, and approval from both the Regional and Federal Medical Councils was required, along with consent from the patient's legal guardian. Post-surgery, the patient demonstrated significant

Table 1. Summary of previous case reports where bilateral anterior capsulotomy and hypothalamotomy for aggressive behavior non-responsive to medication were performed.

Year	Author	Age/Sex	Neuropsychiatric Overview	Procedure Details	Study Conclusions
2015	Santos et al. ¹¹	27 years/Male	Organic delusional disorder (schizophrenic type) and absence epilepsy, with hetero-aggressive behavior	Stimulation at frequencies ranging from 5 Hz to 100 Hz was utilized, followed by a single ablation of the hypothalamus and bilateral ablation of the anterior leg of the internal capsule, both performed at 80 °C for 70 seconds each	No postoperative complications were reported, and aggressive behavior completely ceased for the duration of the two years post-surgery. Seizures and schizophrenia remained unchanged
2017	Borges et al. ¹⁸	16 years/Male	Hypoxia at birth. Severe intellectual disability, hypersexuality, hetero-aggressive behavior and self-injurious behavior	Not reported	No postoperative complications were reported, and the patient's symptoms significantly improved
2018	Lopes et al. ²¹	13 years/Not reported	Severe intellectual disability, associated with hetero-aggressive behavior and self-injurious behavior	Not reported	Post-operative progress and aggression control were satisfactory

improvement, responding to previously ineffective psychiatric medications, with no self-harm episodes over the past three years. Psychosurgery therefore remains a last-resort treatment for self-injurious behavior when all other options have failed, and the patient's life is at risk.

CONCLUSION

Severe self-injurious behavior can be highly debilitating, especially when resistant to conventional treatments. Psychosurgery has evolved significantly since its inception and has become an effective option for such refractory cases, provided that each patient undergoes careful evaluation, and the protocol established by the Medical Council is followed. In this case report, a bilateral stereotactic anterior capsulotomy and posteromedial hypothalamotomy were performed on a patient with severe SIB, ceasing his self-mutilation when combined with previously unsatisfactory psychiatric medications. Further research is needed to deepen our understanding of this surgical approach.

REFERENCES

1. Favazza AR. The coming of age of self-mutilation. *J Nerv Ment Dis.* 1998;186(5):259-68. <http://doi.org/10.1097/00005053-199805000-00001>. PMID:9612442.
2. Anandan S, Wigg CL, Thomas CR, Coffey B. Psychosurgery for self-injurious behavior in Tourette's disorder. *J Child Adolesc Psychopharmacol.* 2004;14(4):531-8. <http://doi.org/10.1089/cap.2004.14.531>. PMID:15662144.
3. Faria MA Jr. Violence, mental illness, and the brain - a brief history of psychosurgery: Part 1 - From trephination to lobotomy. *Surg Neurol Int.* 2013;4(1):49. <http://doi.org/10.4103/2152-7806.110146>. PMID:23646259.
4. Robison RA, Taghva A, Liu CY, Apuzzo ML. Surgery of the mind, mood, and conscious state: an idea in evolution. *World Neurosurg.* 2012;77(5-6):662-86. <http://doi.org/10.1016/j.wneu.2012.03.005>. PMID:22446082.
5. Mashour GA, Walker EE, Martuza RL. Psychosurgery: past, present, and future. *Brain Res Brain Res Rev.* 2005;48(3):409-19. <http://doi.org/10.1016/j.brainresrev.2004.09.002>. PMID:15914249.

6. Moniz E. Prefrontal leucotomy in the treatment of mental disorders. 1937. *Am J Psychiatry*. 1994;151(6, Suppl):236-9. <http://doi.org/10.1176/ajp.151.6.236>. PMID:8192205.
7. Masiero AL. A lobotomia e a leucotomia nos manicômios brasileiros. *Hist Cienc Saude Manguinhos*. 2003;10(2):549-72. <http://doi.org/10.1590/S0104-59702003000200004>. PMID:14567359.
8. Longo PW, Pimenta AM, Arruda J. Lobotomia pré-frontal. Resultados clínicos em hospital privado. *Arq Neuropsiquiatr*. 1949;7(2):126-40. <http://doi.org/10.1590/S0004-282X1949000200002>. PMID:18146411.
9. Yahn M. Sobre a leucotomia pré-frontal de Egas Moniz. *Arq Neuropsiquiatr*. 1946;4(3):213-38. <http://doi.org/10.1590/S0004-282X1946000300001>.
10. Bertolote JM. Egas Moniz: twice a double life. *Arq Neuropsiquiatr*. 2015;73(10):885-6. <http://doi.org/10.1590/0004-282X20150138>. PMID:26465405.
11. Santos R, Mengai A, Silva L, Arruda J. Surgical treatment of aggressive behavior: case report. *J Bras Neurocir*. 2015;25(4):367-72. <http://doi.org/10.22290/jbnc.v25i4.1259>.
12. Gusmão SS, Silveira RL, Arantes A. Stereotatic neurosurgery: a review of its development, principles, technique and applications. *J Bras Neurocir*. 1998;9(2):56-63. <http://doi.org/10.22290/jbnc.v9i2.271>.
13. Pepper J, Hariz M, Zrinzo L. Deep brain stimulation versus anterior capsulotomy for obsessive-compulsive disorder: a review of the literature. *J Neurosurg*. 2015;122(5):1028-37. <http://doi.org/10.3171/2014.11.JNS132618>. PMID:25635480.
14. BRASIL. Lei no 10.216, de 6 de abril de 2001. Dispõe sobre a proteção e os direitos das pessoas portadoras de transtornos mentais e redireciona o modelo assistencial em saúde mental. *Diário Oficial da União*; Brasília; 6 abril 2001.
15. BRASIL. Conselho Federal de Medicina. Resolução CFM nº. 2.057/2013. *Diário Oficial da União*; Brasília; 12 novembro 2013.
16. Patel SR, Aronson JP, Sheth SA, Eskandar EN. Lesion procedures in psychiatric neurosurgery. *World Neurosurg*. 2013;80(3-4):31.e9-16. <http://doi.org/10.1016/j.wneu.2012.11.038>. PMID:23159652.
17. Pepper J, Zrinzo L, Hariz M. Anterior capsulotomy for obsessive-compulsive disorder: a review of old and new literature. *J Neurosurg*. 2019;133(5):1595-604. <http://doi.org/10.3171/2019.4.JNS19275>. PMID:31604328.
18. Borges TJB, Assunção APA, Silva LJ. A importância de múltiplos alvos no tratamento cirúrgico da agressividade refratária. *Rev Med Saude Brasilia*. 2017;6(2):193-202.
19. Teixeira MJ, Guertzenstein EZ, Hamani C, Venturini L, Calazans MEB, Montenegro MC. Cirurgia psiquiátrica para agressividade. *J Bras Neurocir*. 2005;15(3):105-11. <http://doi.org/10.22290/jbnc.v15i3.493>.
20. Sano K, Mayanagi Y. Posteromedial hypothalamotomy in the treatment of violent, aggressive behaviour. *Acta Neurochir Suppl*. 1988;44:145-51. http://doi.org/10.1007/978-3-7091-9005-0_28. PMID:3066130.
21. Lopes MV, Teixeira ML, Pinheiro LS, et al. Neurocirurgia funcional no tratamento da agressividade. *Arquivos Brasileiros de Neurocirurgia: Brazilian Neurosurgery*. 2018;37(S01):1-332. <http://doi.org/10.1055/s-0038-1672660>.
22. Gouveia FV, Hamani C, Fonoff ET, et al. Amygdala and hypothalamus: historical overview with focus on aggression. *Neurosurgery*. 2019;85(1):11-30. <http://doi.org/10.1093/neuros/nyy635>. PMID:30690521.

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abordagem não invasiva no manejo da pressão intracraniana

reduza o **risco da hipertensão intracraniana**
nos **pacientes pré e pós-cirúrgicos**
no **consultório, ambulatório**
ou a **beira-leito**

a tecnologia brain4care:

- Indica se **pacientes** estão sob risco de hipertensão intracraniana
- Auxilia na **proatividade do manejo de edemas** encefálicos
- **Qualifica o diagnóstico e tratamento** de distúrbios como HPN, demências reversíveis e outras hidrocefalias
- **Suporta o processo de verificação e ajuste de válvulas**

contribui para:

- Justificar a **pertinência de neurocirurgias**
- Alta segura e redução do **risco de danos neurológicos secundários** pós-cirúrgicas
- **Reembolso** com códigos **validados pela AMB**

registro da ANVISA ✓
liberada pela FDA ✓

nova resolução da AMB 067/24

Agora o código CBHPM
2.02.02.06-7 pode ser aplicado para
uso hospitalar e ambulatorial.

Referente ao código 2.02.02.06-7:

- A monitorização da pressão intracraniana pode ser realizada de forma:
 - invasiva, com instalação do cateter de PIC em ambiente hospitalar; e
 - não invasiva, realizado no ambiente hospitalar, ambulatorial ou consultório.
- O procedimento de monitorização da pressão intracraniana pode ser associado a outros atendimentos médicos como consultas médicas, visitas, avaliações e/ou acompanhamentos.



Solitary Fibrous Tumor of the Posterior Fossa: case report and literature review

Tumor Fibroso Solitário da Fossa Posterior: relato de caso e revisão da literatura

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ABSTRACT

Solitary fibrous tumor (SFT) is an uncommon intracranial tumor that is classified into grades I, II and III to its aggressiveness. The following case report details the case of a 50-year-old male patient who had been suffering from headaches, nausea, and gait alterations for a period of three months, with a progressive exacerbation of these symptoms. A CT scan of the skull revealed an expansive hyperdense cerebellar lesion on the right. Magnetic resonance imaging revealed a solid lesion with homogeneous gadolinium enhancement, accompanied by a small cystic area, characterized by hyposignal on T1 and heterogeneous hypersignal on T2, suggesting a diagnosis of meningioma. Surgical resection was performed, and the anatomopathological analysis indicated SFT grade I according to the World Health Organization (WHO) classification system. Solitary fibrous tumors are frequently misdiagnosed as meningiomas due to the presence of comparable neuroimaging characteristics. For this reason, it is imperative to employ a meticulous diagnostic approach that incorporates an anatomopathological examination and an immunohistochemical panel to ensure an accurate diagnosis of intracranial neoplasms.

Keywords: Infratentorial neoplasms; Cranial fossa, posterior; Solitary fibrous tumors

RESUMO

O tumor fibroso solitário (TFS) é um tumor intracraniano raro sendo classificado em graus I, II e III, conforme sua agressividade. Relatamos um caso de um paciente masculino, 50 anos, com quadro de cefaleia, náuseas e alteração de marcha há três meses com piora progressiva. A tomografia de crânio evidenciou lesão expansiva hiperdensa cerebelar a direita. A ressonância magnética revelou uma lesão sólida com realce homogêneo pelo gadolínio com pequena área cística, caracterizada por hipossinal em T1 e hipersinal heterogêneo em T2, sugestivo de meningioma. Foi realizada ressecção cirúrgica e o anatomopatológico indicou TFS grau I pela Organização Mundial da Saúde (OMS). O tumor fibroso solitário é frequentemente confundido com o meningioma devido a características de neuroimagem semelhantes. Por esse motivo, a confirmação diagnóstica com anatomopatológico associado a painel imuno-histoquímico é de extrema importância para o diagnóstico preciso de neoplasias intracranianas.

Palavras-Chave: Neoplasia infratentorial; Fossa posterior; Tumor fibroso solitário

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INTRODUCTION

Intracranial solitary fibrous tumors (SFTs) are extra-axial neoplasms that have their origin in mesenchymal cells. It is characterized by its effect on soft tissues of fusocellular lineage, manifesting a biological behavior that exhibits considerable variability. This particular neoplasm is uncommon, representing less than 1% of all primary intracranial neoplasms¹. Its occurrence is not confined to any specific age group; however, TFS demonstrates a predilection for its prevalence between the fourth and fifth decade of life, exhibiting no discernible gender disparity².

The initial description of this condition was provided by Carneiro et al.³ in 1996, and it was subsequently incorporated into the classification of Central Nervous System (CNS) tumors in 2002 by the World Health Organization (WHO)¹. In 2016, TFS was incorporated, along with hemangiopericytoma (HPC), into the classification of non-meningothelial mesenchymal tumors⁴. In 2021, the term “hemangiopericytoma” was discontinued due to new evidence indicating that both TFS and HPC shared the gene fusion of the NAB2 and STAT6 genes. This results in the nuclear expression of the STAT6 protein. The protein, which is associated with other markers, is part of the immunohistochemical profile. This profile is considered the molecular signature of TFS. Therefore, the fifth edition of the International Federation of Societies of Surgical Oncology (IFSO) and the Histiocyte and Plasma Cell Society (HPC) have become a single entity, which is herein referred to as “solitary fibrous tumor” (SFT)⁵. The classification of SFT is based on its aggressiveness, with the tumor graded as either first, second, or third Grade (II, III, or I, respectively)⁴⁻⁷.

On neuroimaging, TFS manifests as a uniform, isolated expansile lesion with clearly delineated borders. Magnetic resonance imaging (MRI) reveals that the signal intensity is isointense on T1 and iso- or hyperintense on T2⁶. Due to its distinctive characteristics, it is frequently misdiagnosed as a meningioma. In such cases, comprehensive anatomical pathology and immunohistochemical analysis are paramount for accurate diagnosis and treatment planning. In this paper, we present a case study of a patient diagnosed with a solitary fibrous tumor that exhibited characteristics consistent with those of a cerebellar convexity meningioma.

CASE PRESENTATION

A 50-year-old male patient presented at the emergency department with headache, nausea, and altered gait that had persisted for a duration of three months. Notably, there had been a progressive exacerbation of these symptoms over the previous 15 days. A thorough examination revealed that the patient was alert and oriented, exhibiting globally preserved strength and sensitivities. The pupils were isochoric and photoreactive, and the extrinsic ocular motricity, facial mimicry, and bilateral dysmetria were symmetrical. However, the patient exhibited slurred speech, kinetic ataxia, and a bilateral plantar flexor skin reflex.

A computed tomography (CT) scan revealed a hyperdense cerebellar lesion in the right lobe, measuring 4.0 x 3.5 centimeters. This lesion was found to contain a 1.0-centimeter cystic component and was observed to exert an adjacent compressive effect. Additionally, the scan revealed a small adjacent hyperostosis of the posterior fossa (Figure 1).

The investigation was supplemented by contrast-enhanced MRI, which revealed a predominantly solid expansive lesion with homogeneous gadolinium enhancement and a small cystic area. The lesion exhibited hyposignal characteristics on T1 and homogeneous hyposignal characteristics on T2, with dimensions measuring approximately 4.1 x 3.6 x 3.4 cm. These findings are indicative of a cerebellar convexity meningioma (Figure 2).

Neurosurgical intervention was indicated for tumor resection. The surgical procedure was performed with the patient in a prone position, utilizing the Concorde position. A bilateral suboccipital craniotomy was performed, with the right side of the procedure focusing on exposure of the right sigmoid sinus and transverse sinuses bilaterally. Dural thickening was identified in the right cerebellar cortex. The patient underwent dural resection adjacent to the expansive lesion and dissection of the tumor through the cleavage plane. This procedure entailed complete macroscopic tumor resection, removal of the dural implantation, and drilling of the bone hyperostosis.

The patient exhibited a favorable postoperative evolution, with no focal neurological deficits. A post-operative CT scan of the skull revealed no evidence of neurosurgical complications (Figure 3).

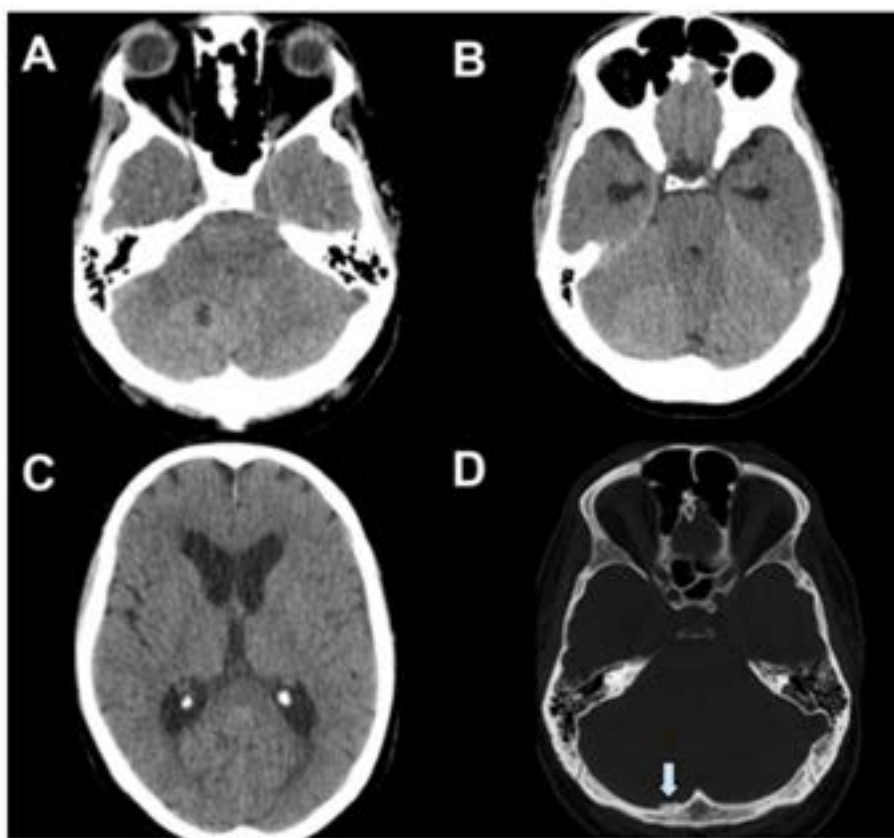


Figure 1. Axial CT scan of the skull. **A** and **B.** Right cerebellar hyperdense expansile lesion. **C.** Ventricular dilatation. **D.** Hyperostosis in the cerebellar cortex on the right.

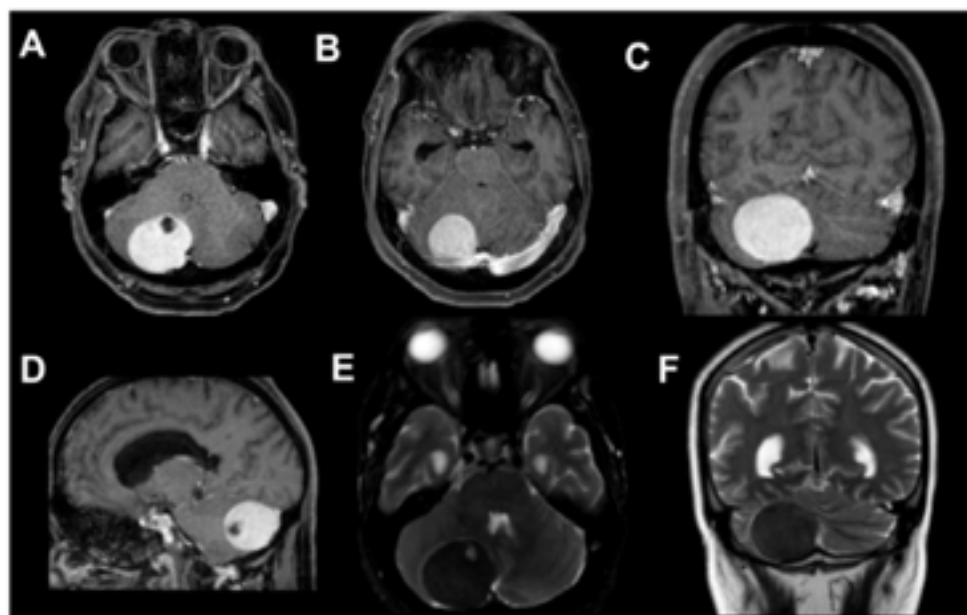


Figure 2. MRI of the skull with gadolinium. **A, B, C** and **D.** contrast-enhanced T1-weighted image: expansile lesion in the right posterior fossa with homogeneous contrast enhancement. **E** and **F.** T2-weighted: presence of cerebrospinal fluid rhyme, indicating an extra-axial lesion. Findings suggestive of medial meningioma of cerebellar convexity.

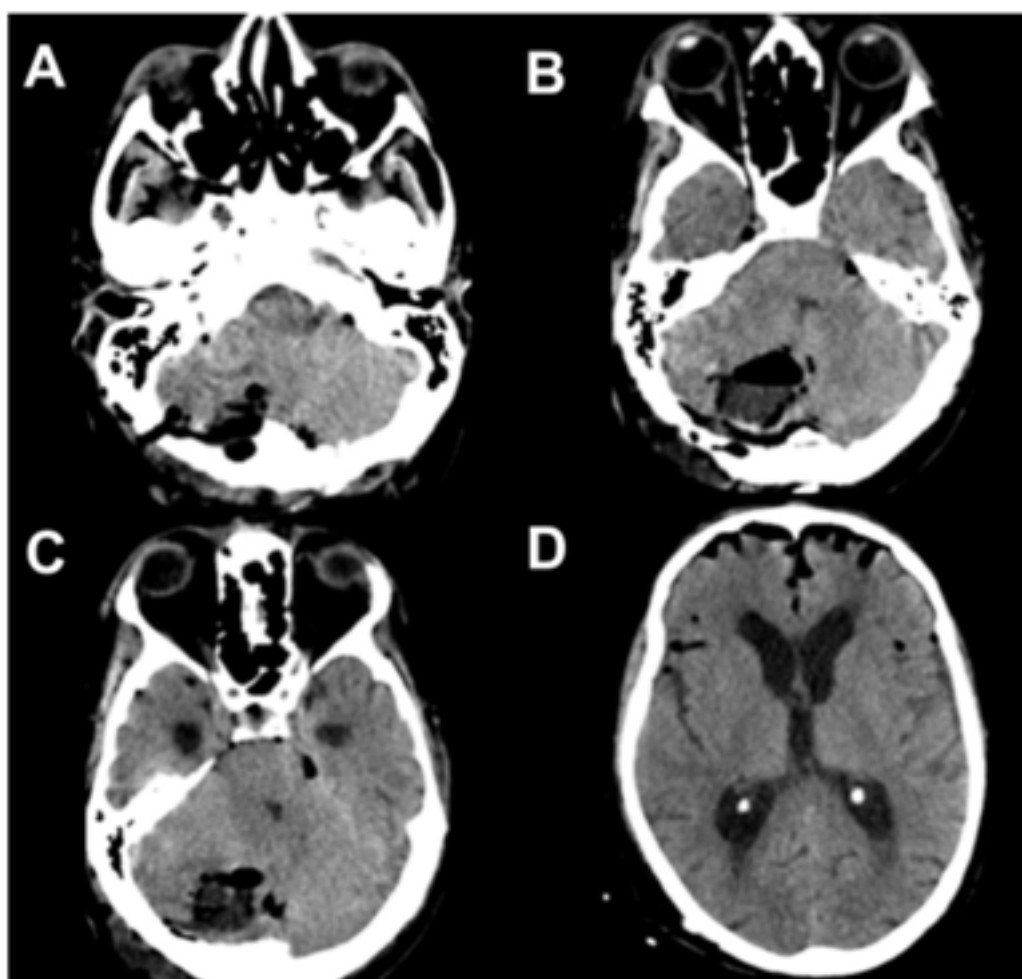


Figure 3. Axial CT scan of the skull. **A, B and C.** Complete resection of the lesion, with no evidence of post-operative complications. **D.** No worsening of ventricular dilation, cortical sulci apparent, no signs of intracranial hypertension.

The anatomopathological analysis of the lesion revealed a mesenchymal neoplasm of spindle cells arranged in bundles, circumscribed, with a low mitotic index, collagenized stroma, and central degenerative changes. These findings support the hypothesis of a solitary fibrous tumor grade I according to the World Health Organization classification. To ensure a comprehensive evaluation and establish a definitive diagnosis, an immunohistochemical panel was conducted. This comprehensive approach enabled the exclusion of other lesions with histopathological characteristics similar to those of TFS. The results of the immunohistochemical panel revealed the following: negative epithelial specific antigen (EMA), positive STAT6, positive podoplanin, Ki-67 < 1%, and positive CD34. The immunohistochemical profile was considered to be compatible with TFS (Figure 4). The patient was discharged from the hospital in stable condition, with no evidence of neurological sequelae.

DISCUSSION

Intracranial solitary fibrous tumor (SFT) is a rare spindle cell neoplasm of mesenchymal origin that accounts for 0.09% of all meningeal neoplasms⁷. Originating in the dura mater and exhibiting histological characteristics analogous to those of meningioma, these tumors have the potential to manifest in the tentorium, the cerebral fossa, and the convex surface of the brain. The most prevalent location of these tumors is in the posterior fossa⁸⁻¹⁰.

The clinical manifestation of this condition varies significantly based on factors such as the tumor's location, dimensions, and degree of invasion¹. Headache is one of the most common symptoms in patients with intracranial lesions, as in the case

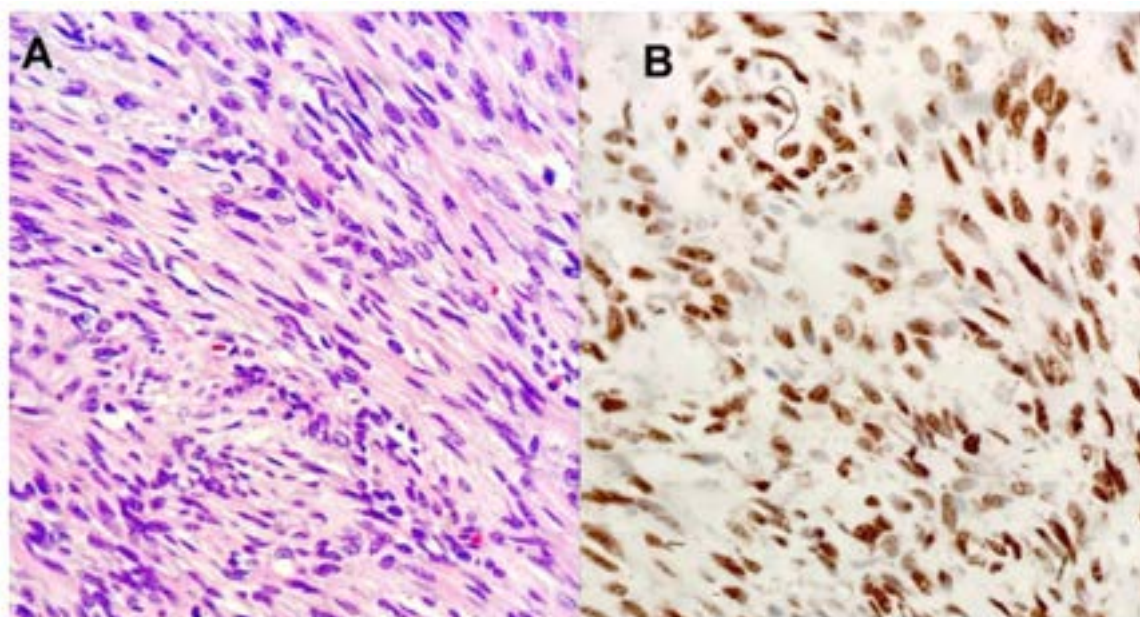


Figure 4. A. Low-grade mesenchymal neoplasm consisting of spindle cells with mild atypia, arranged in bundles/fascicles, with foci of collagenized stroma and rare mitotic figures, indicative of a solitary fibrous tumor (Hematoxylin-eosin, 200x). B. Positive immunohistochemical expression for STAT6, confirming the hypothesis of a solitary fibrous tumor (Ventana Systems, 200x).

reported here, due to the local mass effect caused by the tumor. Furthermore, a multitude of neurological symptoms have been identified in previous studies, including nausea, vomiting, paresis, paresthesia, loss of visual acuity, and seizures¹¹. In the present case, the clinical presentation of ataxia exhibited anatomic-functional correspondence with the site of tumor implantation, which was located in the cerebellar territory.

Diagnostic workup requires imaging studies, including CT and MRI. A solitary fibrous tumor (SFT) is typically characterized by isointense signal on T1-weighted images (T1WIs) and iso- to hyperintense signal on T2-weighted images (T2WIs), exhibiting a uniform enhancement pattern. These tumors frequently manifest well-defined or multilobulated margins, a narrow implantation base, and a more extensive infiltrative component accompanied by apparent parenchymal invasion and bone erosion^{6,12}. In certain instances, they may manifest an irregular pattern of hypo- and hyperintensity on T2-weighted images, giving rise to a distinctive black and white or yin-yang appearance⁸. Furthermore, TFS exhibits a pronounced vascular component, frequently manifesting as the intratumoral empty flow sign. This signifies the presence of vascular channels with low signal intensity across all imaging sequences¹³. Conversely, meningiomas, which are often misdiagnosed as TFS due to their similar neuroimaging characteristics, manifest as homogeneous masses with clearly

delineated borders and intense iso- or hypointensity on non-contrast imaging sequences. Furthermore, meningiomas may exhibit intratumoral calcifications, hyperostosis, and dural tail sign, which are rare findings in TFS^{14,15}.

Additionally, the extensive vascularization supplied by the internal carotid and vertebral arteries or the vertebrobasilar system enables angiotomography to complement the diagnosis⁸. Given the propensity of TFS to develop regional recurrences, as well as distant metastases, multimodal imaging, such as proton emission tomography/computed tomography (PET/CT), has been utilized in the diagnostic evaluation, since it allows metabolic and morphological information to be obtained¹⁶.

Microscopically, the solitary fibrous tumor is composed of monomorphic spindle cells, which tend to group together in fascicles without presenting a specific arrangement. The distinguishing features of these lesions include alternating hypo- and hypercellularity, along with a focal perivascular pattern. As indicated by the World Health Organization (WHO), the classification of degrees of aggressiveness is comprised of three distinct grades. Grade I corresponds to spindle cells that are rich in collagen and with low cellularity. Grade II refers to tumors with less collagen and vascularization in the shape of a “deer

antler.” Grade III includes lesions with five or more mitoses per field of application⁴.

Typically, TFS poses a significant diagnostic challenge, and the integration of immunohistochemical and molecular characteristics is paramount in establishing a precise diagnosis. With regard to the molecular aspect, TFS is currently defined by the presence of the fusion of the NAB2 and STAT6 genes by genomic inversion at the 12q13 locus, an initial event in tumor development¹². The STAT6 protein functions as a transcription factor, while NAB2 serves as a transcriptional repressor¹⁹. As indicated by the extant literature, these proteins are integral to the development of tumors. They modulate the immune response, tumor-induced angiogenesis, and the activation of fibroblasts and collagen synthesis. The NAB2/STAT6 fusion results in the nuclear expression of STAT6, which can be detected by immunohistochemistry. This method has been shown to have excellent specificity and sensitivity for identifying TFS^{13,20}. Furthermore, the expression of STAT6 is a valuable tool in the differential diagnosis of neoplasms with similar histology that do not express this marker¹⁹.

The most characteristic immunohistochemical finding, although not highly specific, associated with solitary fibrous tumors is the expression of CD34, which varies between 81% and 95% among the studies²¹⁻²³. Indeed, in the case presented, the histochemical analysis of the tumor tissue was positive for CD34, which is consistent with the extant literature. However, an exception is observed in highly undifferentiated malignant solitary fibrous tumors, which exhibit an attenuation of CD34 expression, though it remains a significant marker. As indicated by the data presented in Figure 4, there appears to be a direct correlation between the degree of anaplasia of the tumor and the sensitivity of CD34 staining in diagnosing TFS. Specifically, as the degree of anaplasia increases, the sensitivity of CD34 staining decreases²⁴.

The evolution of many TFSs is unpredictable, and they exhibit high recurrence rates, metastases ranging from 10% to 40%, and a lack of efficient, standardized systemic treatments²⁵. The aforementioned characteristics indicate the necessity for data that can accurately predict patient outcomes. This data should include clinical and histopathological variables that can effectively stratify risk²⁵. In this sense, one of the immunohistochemical targets utilized is the cell proliferation marker Ki-67. In the case presented here, Ki-67 was identified in less than 1% of the cells analyzed. The extant evidence suggests a correlation between elevated Ki67 expression and the presence of metastases, the advanced clinical stage of the disease, and the degree of undifferentiation of the tumor cells²⁶. In a similar

vein, the amplification of Ki-67 expression has been shown to be strictly associated with elevated recurrence rates of TFS of the pleura and central nervous system (CNS), particularly when expressed by 5% or more of the tumor cells analyzed²⁶.

As previously discussed, one of the main differential diagnoses of solitary fibrous tumor (SFT) is meningioma, especially the fibrous subtype. The tumor in this case exhibits morphological characteristics similar to the fibrous subtype of meningioma due to (i) the conformation of the spindle cells and (ii) the diffuse fascicular growth pattern. In contrast, atypical and anaplastic meningiomas have been observed to mimic the behavior of malignant TFS, exhibiting a disorganized growth pattern with leaf arrangements, areas of necrosis, high mitosis rates, pleomorphism, and cellular atypia^{27,28}. However, it is noteworthy that meningiomas frequently express epithelial-specific antigen (EMA), a marker that is absent in the majority of solitary fibrous tumors, including the case study in this study. Furthermore, all meningioma subtypes are consistently negative for CD34 and STAT6 in immunohistochemical analyses, in contrast to TFS. The present data underscore the efficacy of immunohistochemical analyses in distinguishing between neoplasms²⁹.

The gold standard treatment for solitary fibrous tumor is surgical resection, which can be complemented with chemotherapy and radiotherapy if necessary. Due to the high rates of recurrence and metastatization, complete excision is preferable⁹. The surgical strategy depends on the location of the tumor. It has been demonstrated that due to the substantial blood supply, preoperative embolization can serve as a valuable instrument to reduce tumor size and perioperative bleeding¹. It is important to note that there is currently a lack of evidence supporting the long-term benefits of radiotherapy. Furthermore, studies conducted on patients who have received chemotherapy are limited in number, making it difficult to assess the efficacy of the treatment¹⁷. Nevertheless, some studies have suggested that gamma knife radiosurgery may be an alternative treatment for solitary fibrous tumors³⁰.

CONCLUSION

The authors present a rare case of a solitary fibrous tumor of the cerebellar convexity. Although TFS is an exceedingly rare condition, it should be considered within the differential diagnoses of neoplasms,

particularly in cases of meningioma. The process of obtaining diagnostic confirmation necessitates the examination of pathological morphology and the implementation of immunohistochemistry.

REFERENCES

- Nhung TH, Minh VL, Lam NL, Lien ND, Duc NM. Malignant intracranial solitary fibrous tumor: a case report and literature review. *Radiol Case Rep.* 2023;18(5):2014-8. <http://doi.org/10.1016/j.radcr.2023.02.064>. PMID:37033688.
- Fargen KM, Opalach KJ, Wakefield D, Jacob RP, Yachnis AT, Lister JR. The central nervous system solitary fibrous tumor: a review of clinical, imaging and pathologic findings among all reported cases from 1996 to 2010. *Clin Neurol Neurosurg.* 2011;113(9):703-10. <http://doi.org/10.1016/j.clineuro.2011.07.024>. PMID:21872387.
- Carneiro SS, Scheithauer BW, Nascimento AG, Hirose T, Davis DH. Solitary fibrous tumor of the meninges: a lesion distinct from fibrous meningioma. A clinicopathologic and immunohistochemical study. *Am J Clin Pathol.* 1996;106(2):217-24. <http://doi.org/10.1093/ajcp/106.2.217>. PMID:8712177.
- Louis DN, Perry A, Reifenberger G, et al. The 2016 World Health Organization classification of tumors of the central nervous system: a summary. *Acta Neuropathol.* 2016;131(6):803-20. <http://doi.org/10.1007/s00401-016-1545-1>. PMID:27157931.
- Louis DN, Perry A, Wesseling P, et al. The 2021 WHO classification of tumors of the central nervous system: a summary. *Neuro-oncol.* 2021;23(8):1231-51. <http://doi.org/10.1093/neuonc/noab106>. PMID:34185076.
- Al Armashi AR, Alkrekshi A, Al Zubaidi A, et al. Grade III solitary fibrous tumor/hemangiopericytoma: an enthralling intracranial tumor - a case report and literature review. *Radiol Case Rep.* 2022;17(10):3792-6. <http://doi.org/10.1016/j.radcr.2022.07.007>. PMID:35965927.
- Gengler C, Guillou L. Solitary fibrous tumour and haemangiopericytoma: evolution of a concept. *Histopathology.* 2006;48(1):63-74. <http://doi.org/10.1111/j.1365-2559.2005.02290.x>. PMID:16359538.
- Sun Z, Li F, Cai X, Jiang Z. Intracranial primary malignant solitary fibrous tumor/hemangiopericytoma masquerading as meningioma: report of a rare case. *Int J Gen Med.* 2020;13:963-7. <http://doi.org/10.2147/IJGM.S279483>. PMID:33149660.
- Cheng L, Ni H, Dai Y. Intracranial solitary fibrous tumor mimicking meningioma: a case report. *Medicine.* 2020;99(50):e23504. <http://doi.org/10.1097/MD.00000000000023504>. PMID:33327290.
- Wu Z, Yang H, Weng D, Ding Y. Rapid recurrence and bilateral lungs, multiple bone metastasis of malignant solitary fibrous tumor of the right occipital lobe: report of a case and review. *Diagn Pathol.* 2015;10(1):91. <http://doi.org/10.1186/s13000-015-0318-9>. PMID:26155787.
- Liu J, Wu S, Zhao K, Wang J, Shu K, Lei T. Clinical features, management, and prognostic factors of intracranial solitary fibrous tumor. *Front Oncol.* 2022;12:915273. <http://doi.org/10.3389/fonc.2022.915273>. PMID:35712477.
- Gubian A, Ganau M, Cebula H, et al. Intracranial solitary fibrous tumors: a heterogeneous entity with an uncertain clinical behavior. *World Neurosurg.* 2019;126:e48-56. <http://doi.org/10.1016/j.wneu.2019.01.142>. PMID:30716501.
- Chikasue T, Uchiyama Y, Tanoue S, Komaki S, Sugita Y, Abe T. Intracranial solitary fibrous tumor/hemangiopericytoma mimicking cystic meningioma: a case report and literature review. *Radiol Case Rep.* 2021;16(7):1637-42. <http://doi.org/10.1016/j.radcr.2021.04.008>. PMID:34007374.
- Buerki RA, Horbinski CM, Kruser T, Horowitz PM, James CD, Lukas RV. An overview of meningiomas. *Future Oncol.* 2018;14(21):2161-77. <http://doi.org/10.2217/fon-2018-0006>. PMID:30084265.
- Watts J, Box G, Galvin A, Brothie P, Trost N, Sutherland T. Magnetic resonance imaging of meningiomas: a pictorial review. *Insights Imaging.* 2014;5(1):113-22. <http://doi.org/10.1007/s13244-013-0302-4>. PMID:24399610.
- Sardaro A, Mammucci P, Pisani AR, et al. Intracranial solitary fibrous tumor: a "new" challenge for PET Radiopharmaceuticals. *J Clin Med.* 2022;11(16):4746. <http://doi.org/10.3390/jcm11164746>. PMID:36012988.
- Wu Z, Yang H, Weng D, Ding Y. Rapid recurrence and bilateral lungs, multiple bone metastasis of malignant solitary fibrous tumor of the right occipital lobe: report of a case and review. *Diagn Pathol.* 2015;10(1):91. <http://doi.org/10.1186/s13000-015-0318-9>. PMID:26155787.
- Kasper E, Lam FC, Anderson M, Boruchow S, Zinn PO, Mahadevan A. "Hitting all the right markers to save a life" Solitary fibrous tumors of the central nervous system: case series and review of the literature. *Surg Neurol Int.* 2012;3(1):83. <http://doi.org/10.4103/2152-7806.99173>. PMID:22937483.
- Doyle LA, Vivero M, Fletcher CD, Mertens F, Hornick JL. Nuclear expression of STAT6 distinguishes solitary fibrous tumor from histologic mimics. *Mod Pathol.* 2014;27(3):390-5. <http://doi.org/10.1038/modpathol.2013.164>. PMID:24030747.
- Schweizer L, Koelsche C, Sahm F, et al. Meningeal hemangiopericytoma and solitary fibrous tumors carry the NAB2-STAT6 fusion and can be diagnosed by nuclear expression of STAT6 protein. *Acta Neuropathol.* 2013;125(5):651-8. <http://doi.org/10.1007/s00401-013-1117-6>. PMID:23575898.
- Bouvier C, Bertucci F, Métellus P, et al. ALDH1 is an immunohistochemical diagnostic marker for solitary fibrous tumours and haemangiopericytomas of the meninges emerging from gene profiling study. *Acta Neuropathol Commun.* 2013;1:10. <http://doi.org/10.1186/2051-5960-1-10>. PMID:24252471.

22. Macagno N, Figarella-Branger D, Mokthari K, et al. Differential diagnosis of meningeal SFT-HPC and meningioma: which immunohistochemical markers should be used? *Sou J Surg Pathol*. 2016;40:270-8. <http://doi.org/10.1097/PAS.0000000000000526>.
23. Ahmad Z, Tariq MU, Din NU. Meningeal solitary fibrous tumor/hemangiopericytoma: emphasizing on STAT 6 immunohistochemistry with a review of literature. *Neurol India*. 2018;66(5):1419-26. <http://doi.org/10.4103/0028-3886.241365>. PMID:30233017.
24. Yamashita D, Suehiro S, Kohno S, et al. Intracranial anaplastic solitary fibrous tumor/hemangiopericytoma: immunohistochemical markers for definitive diagnosis. *Neurosurg Rev*. 2021;44(3):1591-600. <http://doi.org/10.1007/s10143-020-01348-6>. PMID:32671693.
25. Zhang R, Yang Y, Hu C, et al. Comprehensive analysis reveals potential therapeutic targets and an integrated risk stratification model for solitary fibrous tumors. *Nat Commun*. 2023;14(1):7479. <http://doi.org/10.1038/s41467-023-43249-4>. PMID:37980418.
26. Yamamoto Y, Hayashi Y, Murakami I. Recurrence of solitary fibrous tumor/hemangiopericytoma could be predicted by Ki-67 regardless of its origin. *Acta Med Okayama*. 2020;74(4):335-43. PMID:32843765.
27. El-Abtah ME, Murayi R, Lee J, Recinos PF, Kshetry VR. Radiological differentiation between intracranial meningioma and solitary fibrous tumor/hemangiopericytoma: a systematic literature review. *World Neurosurg*. 2023;170:68-83. <http://doi.org/10.1016/j.wneu.2022.11.062>. PMID:36403933.
28. Ohba S, Murayama K, Nishiyama Y, et al. Clinical and radiographic features for differentiating solitary fibrous tumor/hemangiopericytoma from meningioma. *World Neurosurg*. 2019;130:e383-92. <http://doi.org/10.1016/j.wneu.2019.06.094>. PMID:31233926.
29. Tariq MU, Din NU, Abdul-Ghaffar J, Park YK. The many faces of solitary fibrous tumor; diversity of histological features, differential diagnosis and role of molecular studies and surrogate markers in avoiding misdiagnosis and predicting the behavior. *Diagn Pathol*. 2021;16(1):32. <http://doi.org/10.1186/s13000-021-01095-2>. PMID:33879215.
30. Usuda D, Yamada S, Izumida T, et al. Intracranial malignant solitary fibrous tumor metastasized to the chest wall: A case report and review of literature. *World J Clin Cases*. 2020;8(20):4844-52. <http://doi.org/10.12998/wjcc.v8.i20.4844>. PMID:33195652.

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Lipomatous Meningioma: report of a rare variant

Meningioma Lipomatoso: relato de uma variante rara

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Samya Hamad Mehanna² 

ABSTRACT

Lipomatous meningioma is an extremely rare histological variant of meningioma, characterized by cells with peripheral nuclei and clear cytoplasm due to intracellular lipid accumulation. Diagnosis relies on immunohistochemical markers, including epithelial membrane antigen (EMA) and S-100 protein. Clinical presentation and management depend on the tumor's location and size, with headaches and seizures being among the most common symptoms. This subtype predominantly affects women over the age of 55. Given its 17% recurrence rate, gross total resection is the preferred surgical approach. This case study describes the successful surgical intervention in a 48-year-old patient with lipomatous meningioma, contributing to the understanding and management of this rare neoplasm of the Central Nervous System.

Keywords: Intracranial neoplasms; Meningioma; Variant

RESUMO

O meningioma lipomatoso é uma variante histológica extremamente rara do meningioma, caracterizada por células com núcleos periféricos e citoplasma claro devido ao acúmulo intracelular de lipídios. O diagnóstico baseia-se em marcadores imuno-histoquímicos, incluindo o antígeno de membrana epitelial (EMA) e a proteína S-100. A apresentação clínica e o manejo dependem da localização e do tamanho do tumor, sendo cefaleias e convulsões os sintomas mais comuns. Este subtipo afeta predominantemente mulheres com mais de 55 anos. Dado o seu índice de recorrência de 17%, a ressecção total macroscópica é a abordagem cirúrgica preferida. Este estudo de caso descreve a intervenção cirúrgica bem-sucedida em uma paciente de 48 anos com meningioma lipomatoso, contribuindo para a compreensão e o manejo dessa rara neoplasia do sistema nervoso central.

Palavras-Chave: Neoplasias intracranianas; Meningioma; Variante

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INTRODUCTION

Meningiomas comprise a family of neoplasms that are most likely derived from the meningotheial cells of the arachnoid mater (CNS WHO grade 1, 2, or 3). Metaplastic meningioma is a subtype of meningioma characterized by the presence of mesenchymal components within the tumor tissue. These mesenchymal components may include osseous, cartilaginous, lipomatous, myxoid, or xanthomatous elements, either independently or in combination. Despite its designation, the precise mechanisms underlying its pathogenesis remain unclear, and these tumors do not necessarily undergo a true metaplastic transformation^{1,2}.

Lipomatous meningioma is an exceptionally rare variant of metaplastic meningioma, with fewer than 40 cases reported in the literature to date^{3,4} until this date. Morphologically, it is characterized by meningotheial cells with peripherally located nuclei and a cytoplasm rich in lipid-like vacuoles, predominantly composed of triglycerides⁵. Clinical presentation and management depend primarily on the tumor's location and size. Similar to other meningiomas, it shows a marked female predominance, with the majority of cases occurring in patients over 55 years of age⁶.

This study aims to describe the clinical, radiological, surgical, and histopathological aspects of a rare case of lipomatous meningioma in a 48-year-old female patient who underwent surgical resection.

CASE PRESENTATION

A 48-year-old woman, with a history of total thyroidectomy in 2019 for papillary thyroid carcinoma, presented in 2023 with complaints of neck pain, fatigue, and headaches. Her medical history included type 2 Diabetes Mellitus, obesity, a 25-year history of smoking, and hypothyroidism secondary to thyroidectomy. A family history of Central Nervous System (CNS) malignancy was also noted, as her mother had been diagnosed with a CNS tumor. Physical examination revealed no significant abnormalities.

Initial investigations included cervical ultrasound (USG) and computed tomography (CT) of the neck, which incidentally revealed abnormal findings in the skull. Consequently, a magnetic resonance imaging (MRI) of the brain was performed which revealed extra-axial tumor lesions with high spontaneous signal intensity on T1-weighted sequences, suggestive of calcified meningiomas, located at the right parietal convexity (1.9×1.1 cm) and anterior to the temporal pole (1.5×1.0 cm). On magnetic susceptibility-weighted imaging, the lesions predominantly demonstrated low signal intensity (Figure 1). Additionally, faint foci of hyperintensity were observed on T2/FLAIR sequences in the cerebral white matter, though these were nonspecific and without associated mass effect or contrast enhancement. No intraparenchymal expansile process, acute or subacute hemorrhage, hydrocephalus, midline shift, or basal cistern effacement was identified. The ventricular system was of normal

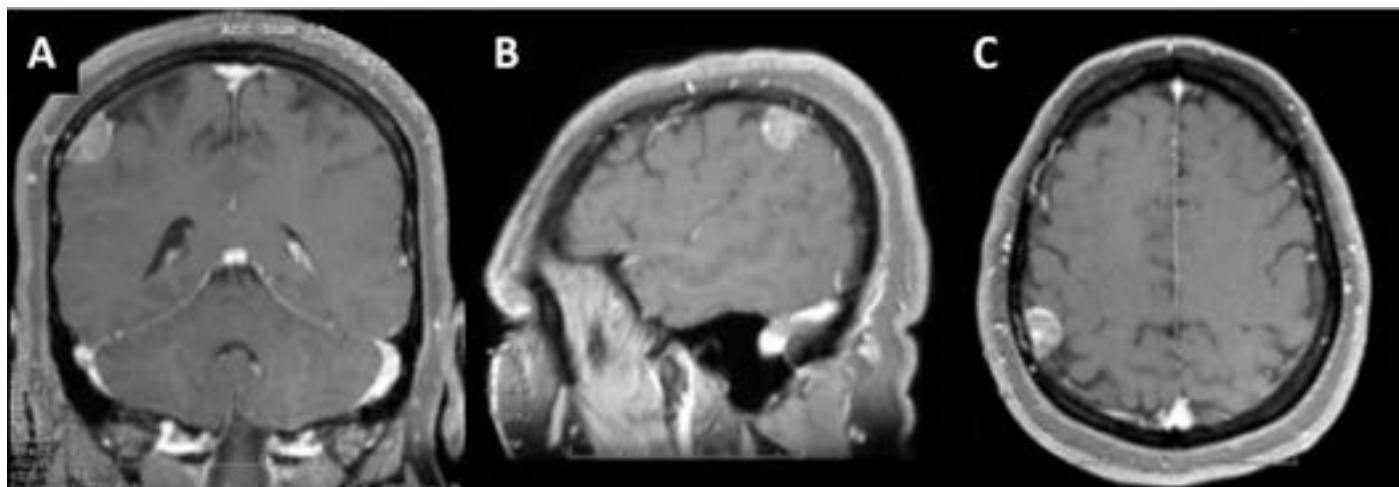


Figure 1. MRI showing extra-axial expansile lesions with high signal intensity on T1 in the right parietal convexity and left temporal pole, possibly calcified/ossified meningiomas, recommending follow-up monitoring. Faint foci of hyperintensity on T2/FLAIR in the white matter of the cerebral hemispheres are nonspecific, possibly related to gliosis or microangiopathy. **A.** Coronal. **B.** Sagittal. **C.** Axial sections.

topography, morphology, and dimensions, and blood flow within the vertebrobasilar and carotid systems was unremarkable.

The patient underwent microsurgical resection of the right parietal convexity meningioma, with no major intraoperative complications. Postoperatively, she was admitted to the Intensive Care Unit (ICU) for observation, where she remained hemodynamically stable without requiring vasopressors or fluid resuscitation. She maintained spontaneous ventilation on room air, with a diuresis output of 900 mL and remained afebrile.

In the microscopic analysis of the lesion, fatty proliferation with classic meningothelial neoplastic cells was observed. The cells have round nuclei containing cytoplasmic vacuoles (Figure 2A and B). The immunohistochemical markers tested in the tumor tissue using the immunoperoxidase technique obtained the following results: weak and focal positive epithelial membrane antigen (EMA), positive progesterone receptor (Figure 2C), positive and multifocal S100 protein, positive MDM2 and negative CDK4.

Thus, the histopathological and immunohistochemical findings confirmed a low-grade lipomatous meningioma (WHO Grade 1, 2021) with a proliferative index (Ki67) of 2%. The patient continues to undergo biannual neurosurgical follow-ups to monitor for potential recurrence.

DISCUSSION

Meningiomas account for one third of all tumors of the Central Nervous System, yet lipomatous meningioma is an extremely rare variant, first reported in 1931 by Bailey and Bucy⁷ and has remained uncommon ever since. This case marks the 40th report of this entity in the literature.

Common symptoms of these types of meningiomas include epileptic seizures and headache, and visual disturbances, hemiparesis, gait instability, and even memory impairment and muscle weakness may also occur. The condition predominantly affects women and is more common in supratentorial sites, such as the frontal and parietal lobes⁶. The most commonly reported symptoms are headaches (common in tumors located in the parietal lobe) and seizures (common in tumors of the frontal and temporal lobes)⁸.

MRI is the main gold standard diagnostic tool for intracranial tumors, including meningioma. The radiological findings described for the lipomatous variant include: extra-axial lesions with high signal intensity on T1-weighted images and low signal intensity on magnetic susceptibility-based sequences that are compatible with the presence of lipomatous tissue, a distinctive feature⁹. However, the presence of calcifications or ossifications,

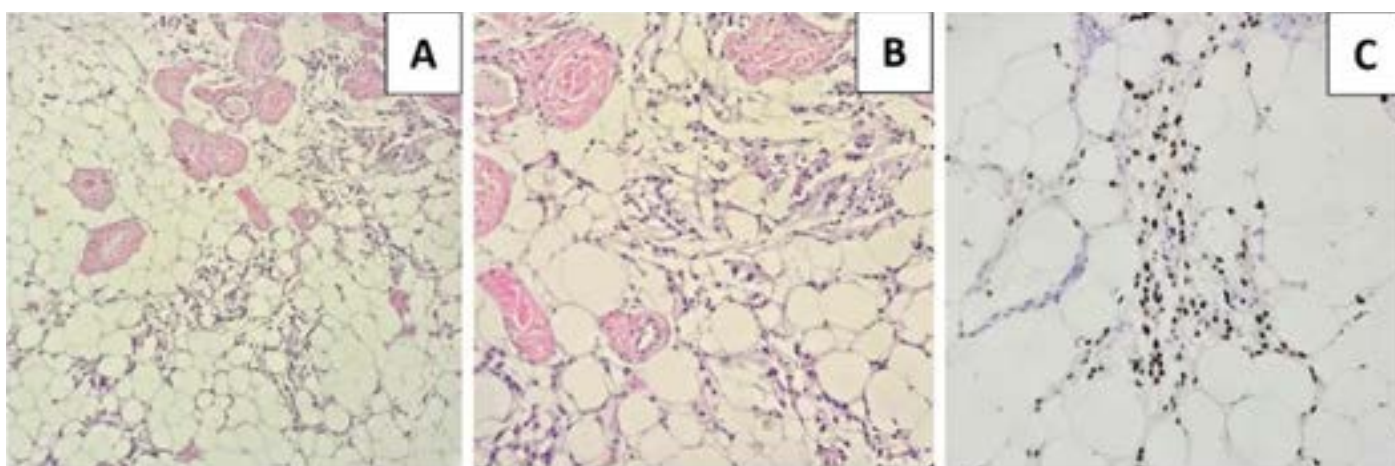


Figure 2. Histopathological and immunohistochemical analysis of the tumor sample. **A** and **B**. Neoplasia composed of clusters of meningothelial cells immersed in abundant adipose tissue, characterized by large optically empty vacuoles corresponding to lipid accumulations, indicating the preservation of the lipid architecture in the sample (Light optical microscopy, Nikon E200, Hematoxylin-eosin staining, 100x and 200x magnifications). **C**. Nuclear positivity for the progesterone receptor in neoplastic cells (Light optical microscopy, Nikon E200, Immunohistochemistry, 400x magnification).

as suggested in the images of the case, reinforces the need for differential diagnosis with other extra-axial lesions, such as hemangiopericytomas or psammomatous meningiomas⁵. The foci of hyperintense signal intensity on T2/FLAIR described in the cerebral white matter are nonspecific, but may be associated with peritumoral gliosis or microangiopathic changes¹⁰.

The microsurgical approach is appropriate for most meningiomas because they are usually located in accessible regions of the central nervous system¹¹. Resection of calcified meningiomas can present technical challenges due to the rigidity of the tumor and the proximity to critical neural and vascular structures, such as the brain, blood vessels, and cranial nerves. However, identification of calcification through imaging tests, both preoperative and intraoperative, allows the surgeon to plan the incision trajectory and the depth of dissection more accurately, with satisfactory results in the vast majority of cases¹².

Detailed analysis of the tumor tissue is essential for the diagnosis of lipomatous meningioma and to rule out other hypotheses. Under the microscope, a proliferation of meningothelial cells is observed, a key characteristic of tumors originating from the meninges, which may be associated with psammomatous bodies in cases where there are calcifications. In addition, the metaplasia of some meningothelial cells in adipose tissue is what gives this tumor variation its name, where lipid tangles are found among the other characteristics present in the tumor mass⁵.

However, this same characteristic of the presence of fat is what can cause doubts in the differentiation between a lipomatous meningioma and other histological types of tumor, such as lipomas, liposarcomas, chordomas, squamous cell tumors or metastatic mucinous carcinomas¹³. Through complementary immunohistochemical examination, it is possible to confirm the diagnosis of meningioma mainly by positivity for epithelial membrane antigen (EMA), somatostatin receptor 2a (SSTR2a) and progesterone receptor (PR), while the lipomatous tumor area expresses the proteins S100 and MDM2. When indolent, the proliferative index assessed by Ki67 is low, less than 5%⁹. Therefore, despite the particular variant, lipomatous meningiomas are classified as Grade 1 by the WHO, demonstrating a low-grade biological behavior, therefore benign¹.

The risk of recurrence of lipomatous meningiomas is 17%, which highlights the importance of complete tumor resection as the treatment of choice for patients with this condition⁶.

Accurate identification through imaging characteristics and immunohistological findings is crucial for accurate diagnosis and determining the most appropriate therapeutic approach. These aspects not only guide initial treatment but also influence long-term monitoring and management strategies, aiming to reduce the recurrence rate and improve clinical outcomes for patients¹².

CONCLUSION

The diagnosis of lipomatous meningioma can be challenging due to the rarity of this neoplastic variant, requiring in-depth immunohistochemical analysis for confirmation. The case description seeks to contribute to the understanding and documentation of an atypical entity, through the detailing of clinical aspects, therapeutic management and prognosis.

REFERENCES

1. Sahm F, Perry A, von Deimling A, et al. Meningioma. In: WHO Classification of Tumours Editorial Board, editors. Central nervous system tumours. 5th ed. Lyon: International Agency for Research on Cancer; 2021. (WHO Classification of Tumours Series; vol. 6).
2. Roncaroli F, Scheithauer BW, Laeng RH, Cenacchi G, Abell-Aleff P, Moschopoulos M. Lipomatous meningioma: a clinicopathologic study of 18 cases with special reference to the issue of metaplasia. *Am J Surg Pathol*. 2001;25(6):769-75. <http://doi.org/10.1097/00000478-200106000-00008>. PMID:11395554.
3. Patel Z, Wang JZ, Merali Z, et al. DNA methylation profiling of a lipomatous meningioma: illustrative case. *J Neurosurg Case Lessons*. 2023;5(6):CASE22518. <http://doi.org/10.3171/CASE22518>. PMID:36748749.
4. Lapolla P, Bruzzaniti P, Zancana G, et al. Visual aura secondary to supratentorial lipomatous meningioma: a rare case report. *Diagnostics*. 2022;12(2):365. <http://doi.org/10.3390/diagnostics12020365>. PMID:35204456.
5. Jaiswal AK, Mehrotra A, Kumar B, et al. Lipomatous meningioma: a study of five cases with brief review of literature. *Neurol India*. 2011;59(1):87-91. <http://doi.org/10.4103/0028-3886.76876>. PMID:21339670.
6. Lapolla P, Familiari P, Zancana G, et al. Lipomatous meningioma: clinical- pathological findings, imaging characterisation and correlations of a rare type of meningioma. *In Vivo*. 2021;35(6):3031-7. <http://doi.org/10.21873/invivo.12598>. PMID:34697134.

7. Bailey P, Bucy PC. The origin and nature of meningeal tumors. *Am J Cancer*. 1931;15:15-54.
8. Gasparinho MG, Ferreira M, Lavrador JP, Livraghi S. Revisiting lipomatous meningioma: a case report and review of an unusual entity. *Int J Surg Pathol*. 2015;23(5):399-403. <http://doi.org/10.1177/1066896915583695>. PMID:25911563.
9. Colnat-Coulbois S, Kremer S, Weinbreck N, Pinelli C, Auque J. Lipomatous meningioma: report of 2 cases and review of the literature. *Surg Neurol*. 2008;69(4):398-402. <http://doi.org/10.1016/j.surneu.2006.11.072>. PMID:17825370.
10. Lim AT, Chandra RV, Trost NM, McKelvie PA, Stuckey SL. Large anterior temporal Virchow-Robin spaces: unique MR imaging features. *Neuroradiology*. 2015;57(5):491-9. <http://doi.org/10.1007/s00234-015-1491-y>. PMID:25614333.
11. Lynch JC, Schiavini H, Bomfim C, Fonseca JF, Pereira C. Microsurgical resection for parasagittal meningiomas with preservation of the parasagittal sinus and excellent neurovascular control. *Arq Neuropsiquiatr*. 2013;71(5):301-6. <http://doi.org/10.1590/0004-282X20130025>. PMID:23689407.
12. Ahmed N, Ferini G, Hossain MATM, et al. Evaluation of surgical cleavage plane by preoperative magnetic resonance imaging findings in adult intracranial meningiomas. *Life*. 2022;12(4):473. <http://doi.org/10.3390/life12040473>. PMID:35454964.
13. Yüksel MO, Gürbüz MS, Tanrıverdi O, Özmen SA. Lipomatous meningioma: a rare subtype of benign metaplastic meningiomas. *J Neurosci Rural Pract*. 2017;8(1):140-2. <http://doi.org/10.4103/0976-3147.193539>. PMID:28149104.

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Remote Cerebellar Hemorrhage: an unusual cause of post-operative deterioration in neurosurgical patients

Hemorragia Cerebelar Remota: uma causa incomum de deterioração pós-operatória em pacientes neurocirúrgicos

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ABSTRACT

Post-operative neurological deterioration in a neurosurgical patient can result from varied causes. In spite of precautions taken to prevent these post-operative complications arising from common risk factors, some rare complications still occur and sometimes attributable to factors remote from the route of surgery. We report two cases of remote cerebellar hemorrhage that were managed in our facility, the first case followed burr hole drainage for subdural hygroma and the other after lumbar spine decompression with inadvertent intra-operative cerebrospinal spinal fluid leakage. Both patients had neurologic deterioration within 72 hours of surgery and brain computed tomography scans in both patients revealed cerebellar hemorrhage.

Keywords: Spontaneous cerebellar hemorrhage; Post-operative cerebellar bleeding; Cerebrospinal fluid leak

RESUMO

A deterioração neurológica pós-operatória em pacientes neurocirúrgicos pode resultar de causas variadas. Apesar das precauções tomadas para prevenir complicações pós-operatórias decorrentes de fatores de risco comuns, algumas complicações raras ainda ocorrem e, por vezes, são atribuíveis a fatores distantes do local da cirurgia. Relatamos dois casos de hemorragia cerebelar remota que foram gerenciados em nossa instituição, o primeiro caso após drenagem de higroma subdural por trepanação e o outro após descompressão da coluna lombar com vazamento inadvertido de líquido cefalorraquidiano durante a operação. Ambos os pacientes apresentaram deterioração neurológica dentro de 72 horas após a cirurgia e exames de tomografia computadorizada cerebral revelaram hemorragia cerebelar.

Palavras-Chave: Hemorragia cerebelar espontânea; Sangramento cerebelar pós-operatório; Vazamento de líquido cefalorraquidiano

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INTRODUCTION

Post-operative neurological deterioration (POND) in a neurosurgical patient can result from varied causes and mechanisms. In spite of precautions taken to prevent these post-operative complications arising from common risk factors, some rare complications still occur and are sometimes attributable to factors remote from the route of surgery. When deterioration occurs following operative approaches that do not involve the posterior fossa, cerebellar hemorrhage is hardly considered as a possible reason due to its location and rarity. This remote cerebellar hemorrhage (RCH) has been noted in procedures involving the brain or the spine^{1,2}. Chaddock reported the first case of RCH following cervical laminectomy with inadvertent cerebrospinal fluid (CSF) leak in 1981³. Since then, only a few cases have been reported in the literature globally, with none reported from sub-Saharan Africa. Since the inception of Memfys hospital for Neurosurgery in the year 2002, with over 40,108 patients managed and 4,872 cases of brain and spine surgical procedures, only two cases of RCH have been recorded. The aim of this paper is to bring the awareness of this problem to the public, discuss the possible mechanisms and highlight the team's management of these cases.

CLINICAL CASE PRESENTATION

Case 1: A 75-year-old right-handed man who presented with a two-day history of weakness of right upper and lower limbs and several episodes of focal seizure involving the right side of the body of seven-hour duration. He was involved in a road traffic accident three weeks earlier with associated loss of consciousness which was subsequently regained about 24 hours later. He is not a known hypertensive or diabetic and medication review revealed no use of anticoagulant or anti-platelet medication.

Physical examination revealed an elderly man with Glasgow coma score (GCS) of 11 (E4V1M6). Vital signs were Pulse- 78/min, BP- 160/90mmHg, RR- 22/min. Both pupils were 2.5mm round and reactive to light briskly. He had right fascioplegia (upper motor neuron type) and right hemiparesis (0/5 in the upper limb and 2/5 in the lower limb using the Medical Research Council (MRC) scale. Preoperative clotting profile was normal (PT- 12s, INR- 0.92) and platelet count was adequate (205,000 cells/uL). Brain

computed tomography (CT) scan revealed right frontoparietal subdural hygroma with no evidence of posterior fossa bleed (Figure 1 A-B).

Patient had a bifrontal burr hole with drainage of subdural hygroma and subgaleal drain insertion. The intraoperative finding was that of xanthochromic CSF under pressure. Two days post-surgery, GCS improved to E4V4M6. Subgaleal drain was significantly active, draining 650mls of blood-tinged CSF over three days. He deteriorated on third day post-surgery with a 5-point drop in GCS to- E2V2M5 (9/15). Emergency brain CT scan revealed right hemispheric cerebellar hemorrhage- (Figure 2A). Search for possible cause of cerebellar hemorrhage with brain magnetic resonance imaging (MRI) and angiography did not reveal any tumor or vascular malformation.

The subgaleal drain was removed and he was managed conservatively with close neuro-vital signs monitoring. He made a progressive neurological improvement with 5-point gain in GCS - E4V4M6 (14/15) and the right hemiparesis improved to the power of 4/5. However, he still had a residual intention tremor of the right upper limb, past pointing and dysarthria. A repeat check brain CT scan done on the 16th post-operative day revealed a resolving right cerebellar hemorrhage (Figure 2B). He was subsequently discharged home on the 19th postoperative day for follow up at the outpatient clinic.

Case 2

A 79-year-old man with lumbar degenerative spine disease and symptomatic lumbar canal stenosis. He presented with radicular low back pain and weakness of both lower limbs but no sphincteric dysfunction. He had cervical decompression two years earlier. He is a known hypertensive with satisfactory blood pressure control on anti-hypertensives. He had no symptoms referable to the intracranial compartment. At physical examination, he was an elderly man with GCS of 15 and intact higher cortical function. Blood pressure was 130/80mmHg, pre-operative electrocardiogram and chest radiograph were normal. He had quadriplegia (power ranged between grades 4 and 5 in different muscle groups in both upper limbs but between grades 1 and 3 in both lower limbs according MRC scale).

Pre-operative clotting profile result was normal (PT- 12 seconds, PTTK- 29 seconds, INR- 0.96) and platelet count was 346,000cells/uL. Pre-operative MRI scanogram revealed normal findings in the brain supratentorial and infratentorial compartments (Figure 3A). He had a lumbar decompressive

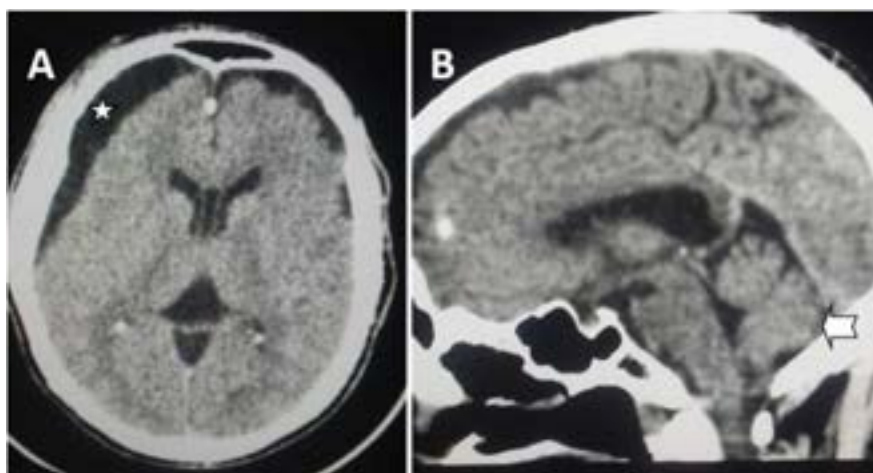


Figure 1. A. Pre-operative Brain CT scan showing bilateral subdural hygroma (star). B. Sagittal view showing a normal cerebellum (arrow).

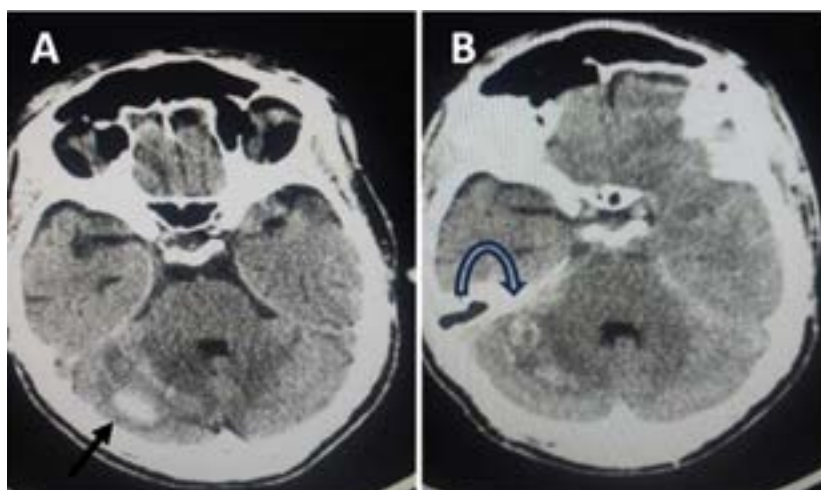


Figure 2. A. Post-operative Brain CT scan showing a bleed in the right cerebellar hemisphere (black arrowhead). B. 16th day post-operative axial brain CT showing resolving cerebellar hematoma (curved arrow)

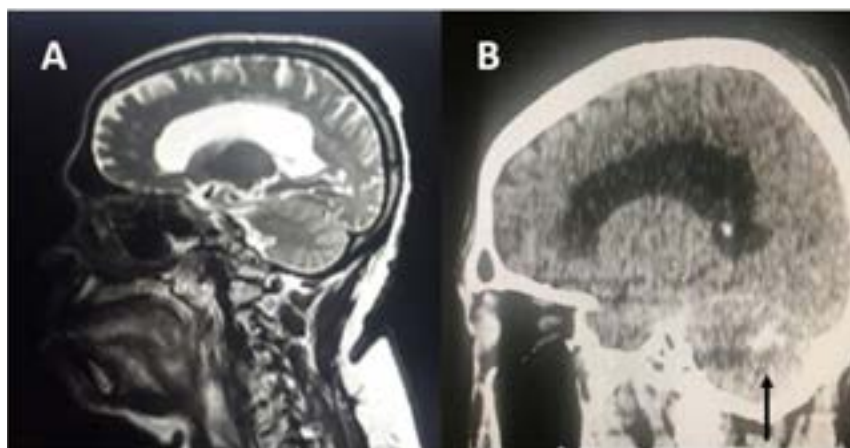


Figure 3. A. Pre-operative Brain MRI sagittal view (from scout T2) with normal posterior fossa. B. Brain CT sagittal view showing cerebellar hemispheric bleed (black straight arrow).

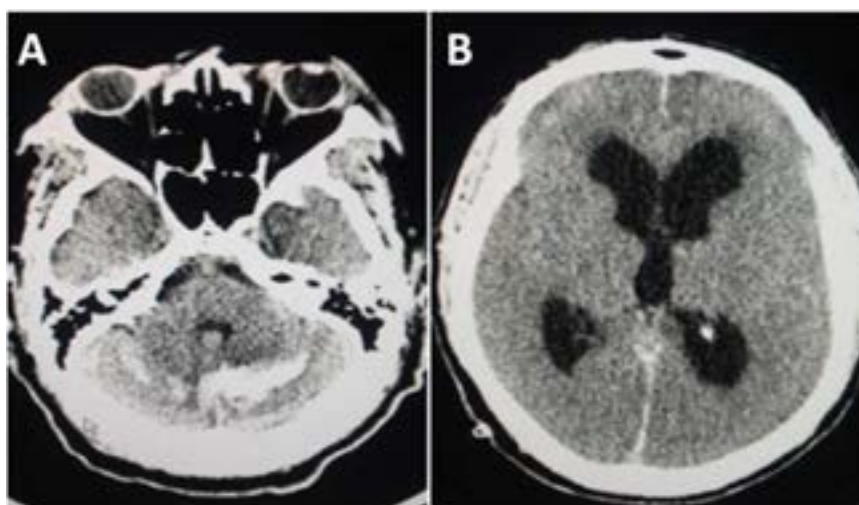


Figure 4. A. Brain CT axial view showing bihemispheric extent of the cerebellar bleed. B. Brain CT Axial view showing the resultant triventriculomegaly.

laminectomy (interlaminar laminectomy, right L2-L5 and left L3-L5) under general anesthesia. Intra-operatively, blood pressure was well controlled, which ranged from 120/70 mmHg to 110/80 mmHg. He had an inadvertent intra-operative dura tear with CSF leak which was repaired with vicryl 4/0 suture. Recovery from general anesthesia was uneventful. However, about three hours post-surgery, he had a witnessed cardiac arrest from which he was promptly resuscitated. Brain CT scan revealed cerebellar hemorrhage (Figures 3B and 4A). He was then transferred to the intensive care unit, intubated and mechanically ventilated with close neuro-vital sign monitoring. Subsequently, features of raised intracranial pressure (systolic hypertension and bradycardia) necessitated a repeat brain CT scan on the fourth day post-surgery which revealed acute hydrocephalus (Figure 4B). He subsequently had a right frontal external ventricular drain. He made some improvement and was subsequently transferred to the ward.

DISCUSSION

Remote cerebellar hemorrhage (RCH) is a rare form of presentation of spontaneous intracerebellar hemorrhage. In our experience over a twenty-year period, the incidence of RCH following chronic subdural ematoma and lumbar spine decompression surgeries are 0.68% and 0.29%, respectively.

Commonly, cerebellar hemorrhage occurs spontaneously due to poorly treated hypertension, amyloid angiopathy, tumors, coagulation disorders and vascular lesions such as aneurysms and arteriovenous malformations¹. Cerebellar hemorrhage could also occur iatrogenically following a posterior fossa operation as a tumor bed bleed. Strict surgical technique such as meticulous hemostasis at the surgical site is expected to reduce post-operative posterior fossa hematoma. However, due to its rarity RCH is not a concept that many surgeons are aware of and as such, not considered and sought for in a post-operative deteriorating neurosurgical patient. Literature search carefully done in sub-Saharan Africa did not record any previous documentation of this subject. Even in global literature, the frequency is still low, with an estimated incidence of 0.08-0.26% following CSF leak in lumbar spine or cranial surgery^{1,4,5}.

The risk factors for this condition are not very well established but advancing age appears to be a remarkable factor as observed in the cases in this report. This is also the trend in the literature⁵⁻⁸. The index cases did not have chronic diseases that cause vasculopathy, there was no history of hematological disorders and the clotting parameters were normal. This relationship with advancing age may be linked to degenerative changes of the vessels in response to sudden change in pressure dynamics along the neural axis. For this reason, the team usually avoids multiple burr holes during drainage of chronic subdural collections in the elderly. We also avoid aggressive on-table aspiration and wash out of fluid in chronic subdural surgery for the elderly.

Another risk factor for RCH is rapid loss of CSF which was observed in both patients we managed. Literature had reported very low incidence of RCH in chronic subdural hematoma drainage surgeries that are not complicated by massive CSF leak^{8,9}.

In reality therefore, the risk of RCH is expected to be high in massive fluid shifts of the milieu interior bathing the brain and spinal cord. Literature has consistently reported the relationship of occurrence of RCH with neurological procedures that had a high volume of cerebrospinal fluid (CSF) loss, as also observed in this case report^{4,5-8}.

The pathomechanism of CSF leak causing RCH is not known for sure. However, suggested mechanisms include a hemodynamic theory as a result of sudden intracranial hypotension (resulting from excessive and rapid CSF loss). This leads to increased transmural pressure resulting in the rupture of posterior fossa bridging veins^{1,2,10,11}. This typical venous source of the hemorrhage is supported by reports of negative angiographic findings in most cases as well as the Zebra pattern on radio-imaging which represents free blood along the cerebellar folia^{2,12}. Another possible mechanism is the mechanical theory from cerebellar sagging which results in stretching and tearing of superior cerebellar veins².

Some researchers have argued that RCH occurs when other mechanisms, such as jugular venous occlusion following extreme neck rotation during positioning, spikes in intraoperative blood pressure, anticoagulant and anti-platelet medication interact simultaneously¹.

About 50% of POND cases attributable to RCH has been reported to occur within the first 24 hours post-surgery as noted in one of our cases¹³. As such, the possibility of RCH should be entertained after procedures with large volume or rapid CSF loss. In the same manner, this unexpected event should constitute one of the points to discuss during counseling of elderly patients especially going for lumbar spine decompression surgery as per the rare chance of cerebellar stroke in the event of CSF leak, although the region anatomically is remote from the site of surgery.

On the one hand, one might have thought that cardiac arrest and the subsequent resuscitative measures might have led to the cerebellar hemorrhage in our second patient. Although arguable, we propose that the cardiac arrest witnessed in this index patient may be due to the cerebellar hemorrhage, as the patient had recovered uneventfully from anesthesia. As support for this idea, Agrawal et al.¹⁴ had reported that intracranial hemorrhage (including cerebellar hemorrhage) is a

possible cause of sudden cardiac arrest of non-cardiac origin through catecholamine-surge induced cardiac stunning¹⁴.

Without a high index of suspicion and neuroimaging, management effort after POND is usually directed at metabolic, medication and circulatory causes especially in centers where the distance to CT scan is significant for fear of patient deterioration en-route the scan room. This case report recommends early scan as soon as possible in POND in order to rule out condition such as RCH. It seems that the prognosis of this condition depends on the volume of hematoma and the associated medical conditions like the presence of cardiac arrest. However, the risk of irreversible deterioration from brainstem involvement may be low in RCH on a background of small hematoma volume and age-related cerebellar atrophy in the elderly^{13,15}. However, it is still necessary to look out for the occasional patient with associated brainstem compression which may require a more aggressive management approach.

CONCLUSION

Surgeons should be aware of this rare complication and make an attempt to mention this in pre-operative counseling as well as promptly request for neuroimaging if there are clinical findings suggestive of neurological decline post procedure with associated excessive CSF leak/drainage. This will prevent misdiagnosis and help expedite appropriate patient stratification and management.

Also, given the added morbidity of RCH, it is important to stress the value of meticulous attempts during and after surgery to prevent uncontrolled massive CSF loss.

REFERENCES

1. Cevik B, Kirbas I, Cakir B, Akin K, Teksam M. Remote cerebellar hemorrhage after lumbar spinal surgery. *Eur J Radiol.* 2009;70(1):7-9. <http://doi.org/10.1016/j.ejrad.2008.01.004>. PMID:18294795.

2. Brockmann MA, Groden C. Remote cerebellar hemorrhage: a review. *Cerebellum*. 2006;5(1):64-8. <http://doi.org/10.1080/14734220500521032>. PMID:16527766.
3. Chadduck WM. Cerebellar hemorrhage complicating cervical laminectomy. *Neurosurgery*. 1981;9(2):185-9. <http://doi.org/10.1227/00006123-198108000-00016>. PMID:7266820.
4. Floman Y, Millgram MA, Ashkenazi E, Rand N. Remote cerebellar hemorrhage complicating unintended durotomy in lumbar spine surgery. *Int J Spine Surg*. 2015;9(9):29. <http://doi.org/10.14444/2029>. PMID:26273547.
5. Huang PH, Wu JC, Cheng H, Shih YH, Huang WC. Remote cerebellar hemorrhage after cervical spinal surgery. *J Chin Med Assoc*. 2013;76(10):593-8. <http://doi.org/10.1016/j.jcma.2013.02.006>. PMID:23746536.
6. Di L, Wei G, Eichberg DG, Komotar RJ, Ivan M. Remote cerebellar hemorrhage associated with intra-operative cerebrospinal fluid leak: a report of two rare case presentations and review of the literature. *Cureus*. 2020;12(12):e12082. <http://doi.org/10.7759/cureus.12082>. PMID:33489500.
7. Kintala S, Jiao K, Ankam A, Paramore CG. Cerebellar hemorrhage and spinal fluid overdrainage with tonsillar herniation following spine surgery. *Cureus*. 2020;12(9):e10418. <http://doi.org/10.7759/cureus.10418>. PMID:33062533.
8. Kaneshiro Y, Yamauchi S, Urano Y, Murata K. Remote hemorrhage after burr-hole surgery for chronic subdural hematoma: a report of two cases. *Surg Neurol Int*. 2019;10(1):18. http://doi.org/10.4103/sni.sni_108_18. PMID:31123625.
9. Chang SH, Yang S-H, Son BC. Cerebellar hemorrhage after burr hole drainage of supratentorial chronic subdural hematoma. *J Korean Neurosurg*. 2009;15(4):592-6. <http://doi.org/10.3340/jkns.2009.46.6.592>. PMID:20062580.
10. Honegger J, Zentner J, Spreer J, Carmona H, Schulze-Bonhage A. Cerebellar hemorrhage arising postoperatively as a complication of supratentorial surgery: a retrospective study. *J Neurosurg*. 2002;96(2):248-54. <http://doi.org/10.3171/jns.2002.96.2.0248>. PMID:11838798.
11. König A, Laas R, Herrmann HD. Cerebellar haemorrhage as a complication after supratentorial craniotomy. *Acta Neurochir*. 1987;88(3-4):104-8. <http://doi.org/10.1007/BF01404145>. PMID:3687495.
12. Bishokarma S, Shrestha S, Devkota UP, et al. Remote cerebellar haemorrhage after surgery for craniopharyngioma: a case report. *Nepal J Neurosci*. 2018;15(2):45-8. <http://doi.org/10.3126/njn.v15i2.20995>.
13. Worm PV, Dalla-Corte A, Brasil AVB, et al. Cerebellar hemorrhage as a complication of spine surgery. *Surg Neurol Int*. 2019;10(85):85. <http://doi.org/10.25259/SNI-121-2019>. PMID:31528423.
14. Agrawal A, Cardinale M, Frenia D, Mukherjee A. Cerebellar Haemorrhage Leading to Sudden Cardiac Arrest. *J Crit Care Med*. 2020;6(1):71-3. <http://doi.org/10.2478/jccm-2020-0007>. PMID:32104734.
15. Sturiale CL, Rossetto M, Ermani M, et al. Remote cerebellar hemorrhage after supratentorial procedures (part 1): a systematic review. *Neurosurg Rev*. 2016;39(4):565-73. <http://doi.org/10.1007/s10143-015-0691-6>. PMID:26846668.

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Intracranial Infectious Aneurysm Secondary to Cerebral Empyema in a Pediatric Patient: case report and systematic review

Aneurisma Infecioso Intracraniano Secundário a Empiema Cerebral em Paciente Pediátrico: relato de caso e revisão sistemática

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ABSTRACT

Intracranial Infectious Aneurysms (IIAs) arise from inflammation of the arterial wall due to bacterial or fungal infections. Representing 0.7%–6.5% of cerebral aneurysms, they are rare in children and often multiple and small. Early antibiotic therapy is essential, yet treatment guidelines remain unclear. We report the case of an 11-year-old with a mycotic aneurysm secondary to cerebral empyema and bacterial meningoenitis, which resolved with spontaneous thrombosis and no neurological deficits. A systematic review following PRISMA guidelines identified 67 relevant studies from 2,604 screened. Data from these cases revealed a male predominance (66.2%) and mean age of 41 years. Headache (37%), visual disturbances (27.2%), and neuromuscular symptoms (22.2%) were common. Bacteria accounted for 86.4% of infections, especially *Streptococcus* spp. (34.4%). Aneurysms most frequently involved the middle cerebral artery (53.3%), slightly more on the left (53.4%). Surgical treatment was performed in 77.5% of cases, including endovascular approaches in 58%. Full symptom resolution occurred in 51.4% of patients. Overall mortality was 20%, increasing to 25.4% with aneurysm rupture. These findings underscore the importance of early infection control - especially endocarditis - and consideration of IIAs in the differential diagnosis of spontaneous subarachnoid hemorrhage.

Keywords: Intracranial infectious aneurysm; Cerebral empyema; Pediatric patient; Mycotic aneurysm; Case report; Systematic review

RESUMO

Aneurismas Infeciosos Intracranianos (AIIs) surgem da inflamação da parede arterial causada por infecções bacterianas ou fúngicas. Representando 0,7%–6,5% dos aneurismas cerebrais, são raros em crianças e geralmente múltiplos e de pequeno calibre. A antibioticoterapia precoce é essencial, embora ainda não existam diretrizes claras de tratamento. Relatamos o caso de uma criança de 11 anos com aneurisma micótico secundário a empiema cerebral e meningoenite bacteriana, que evoluiu com trombose espontânea e sem déficits neurológicos. Uma revisão sistemática conduzida conforme as diretrizes PRISMA identificou 67 estudos relevantes dentre 2.604 triados. Os dados desses casos revelaram predominância do sexo masculino (66,2%) e idade média de 41 anos. Cefaleia (37%), distúrbios visuais (27,2%) e sintomas neuromusculares (22,2%) foram comuns. As bactérias foram responsáveis por 86,4% das infecções, especialmente *Streptococcus* spp. (34,4%). Os aneurismas afetaram com mais frequência a artéria cerebral média (53,3%), discretamente mais à esquerda (53,4%). Tratamento cirúrgico foi realizado em 77,5% dos casos, incluindo abordagem endovascular em 58%. A resolução completa dos sintomas ocorreu em 51,4% dos pacientes. A mortalidade geral foi de 20%, aumentando para 25,4% nos casos com ruptura aneurismática. Esses achados

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reforçam a importância do controle precoce da infecção - especialmente endocardite - e da inclusão dos AIIs no diagnóstico diferencial da hemorragia subaracnoidea espontânea.

Palavras-Chave: *Aneurisma infeccioso intracraniano; Empiema cerebral; Paciente pediátrico; Aneurisma micótico; Relato de caso; Revisão sistemática*

INTRODUCTION

Intracranial Infectious Aneurysms (IIAs) are conditions resulting from an acute inflammatory reaction of the tunica adventitia, which spreads to the muscular layer, causing its weakening and leading to arterial dilation¹⁻³. They can arise from the infection of a normal arterial wall or a pre-existing aneurysm. However, due to their infectious pathophysiology, they are considered pseudoaneurysms^{1,4,5}. IIAs involve a variety of etiological agents, predominantly bacterial (72.8%), such as *Streptococcus* spp. and *Staphylococcus* spp., followed by fungal infections, including *Aspergillus* sp.^{3,6,7}.

IIAs are typically a rare complication of infectious endocarditis but can also result from other causes of bacteremia, such as cavernous sinus thrombophlebitis, bacterial meningitis, subdural empyema, or orbital cellulitis^{8,9}. They tend to manifest as multiple and smaller aneurysms, accounting for 0.7% to 6.5% of all cerebral aneurysms, and are less common in the pediatric population^{10,11}.

IIAs are also known as “mycotic aneurysms,” a term introduced in 1885 by Sir William Osler to describe a mushroom-shaped aneurysm resulting from subacute bacterial endocarditis. Today, the preferred term is infectious aneurysm, which encompasses aneurysms with diverse infectious etiologies⁷.

Although early diagnosis and treatment are crucial for achieving better outcomes in patients with IIAs, there is still no established therapeutic guideline due to the lack of studies and evidence addressing the specificities of each case^{12,13}. However, most strategies involve antibiotic therapy, either alone or combined with microsurgical or endovascular interventions^{11,12}. The treatment modality should consider factors such as aneurysm size and location, rupture status, presence of mass effect, and vessel fragility¹².

This article aims to report a rare case of IIA in a pediatric patient secondary to cerebral empyema and bacterial meningoencephalitis, as well as to perform a systematic review of the literature on IIAs.

The goal is to describe the main etiological agents, clinical features, therapeutic approaches, and outcomes associated with this condition, contributing to its early recognition and proper management.

METHODS

The protocol of the present study is in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. Population: Adults and children, both sexes, worldwide. Exposure: Diagnosed intracranial infectious aneurysm. Comparison: Age, sex, diagnostic interval, clinical characteristics, diagnosis, radiological features of the aneurysm, and treatment. Outcomes: Follow-up and mortality results.

Search strategy and data sources

A systematic search in the MEDLINE electronic database was conducted using the PubMed MeSH Advanced Search Builder tool, employing the following search commands: (((intracranial) OR (cereb*) OR (brain)) AND ((infect*) OR (mycotic) OR (bacterial))) AND (aneurysm*). The search was performed to identify studies that reported clinical cases of intracranial infectious aneurysms in children and adults. The reference lists of identified studies were reviewed to locate additional relevant case reports.

Study selection

Four reviewers independently screened the titles and abstracts of all citations for eligibility and retrieved those meeting the inclusion criteria. If the information available in the abstract was insufficient to determine eligibility, the full article was retrieved for review. Discrepancies were resolved by consensus among the reviewers when necessary. The reviewers individually analyzed all the articles found. Studies reporting information on children and adults of both sexes with intracranial infectious aneurysms, dated within the last 10 years, were included. Studies were excluded if the full text in English or Portuguese was unavailable, if original data were not reported (review articles), or if intracranial

infectious aneurysm was not the primary focus of the study. A total of 2,604 studies were identified, of which 67 had their data effectively collected and analyzed (Figure 1).

Data extraction

A table was structured to standardize the identification and extraction of data from the selected case reports (Table 1). The following data were collected: age, sex, initial neurological presentation, associated etiological agent, radiological properties, clinical and surgical treatment, symptom remission, and clinical outcome. Case reports lacking the necessary information were excluded from data analysis for statistical purposes.

CASE PRESENTATION

An 11-year-old patient presented to the emergency department with symptoms of vomiting and difficulty moving the lower limbs. He was treated with antiemetics and oral hydration. Six days later, the condition progressed with difficulty ambulating, fever, and drowsiness. Infectious screening tests were requested, and empirical treatment with ceftriaxone was initiated. Due to clinical worsening, he was referred to a tertiary pediatric hospital after seven days, where he was evaluated by the pediatrics and neurosurgery teams. During the evaluation, the patient reported

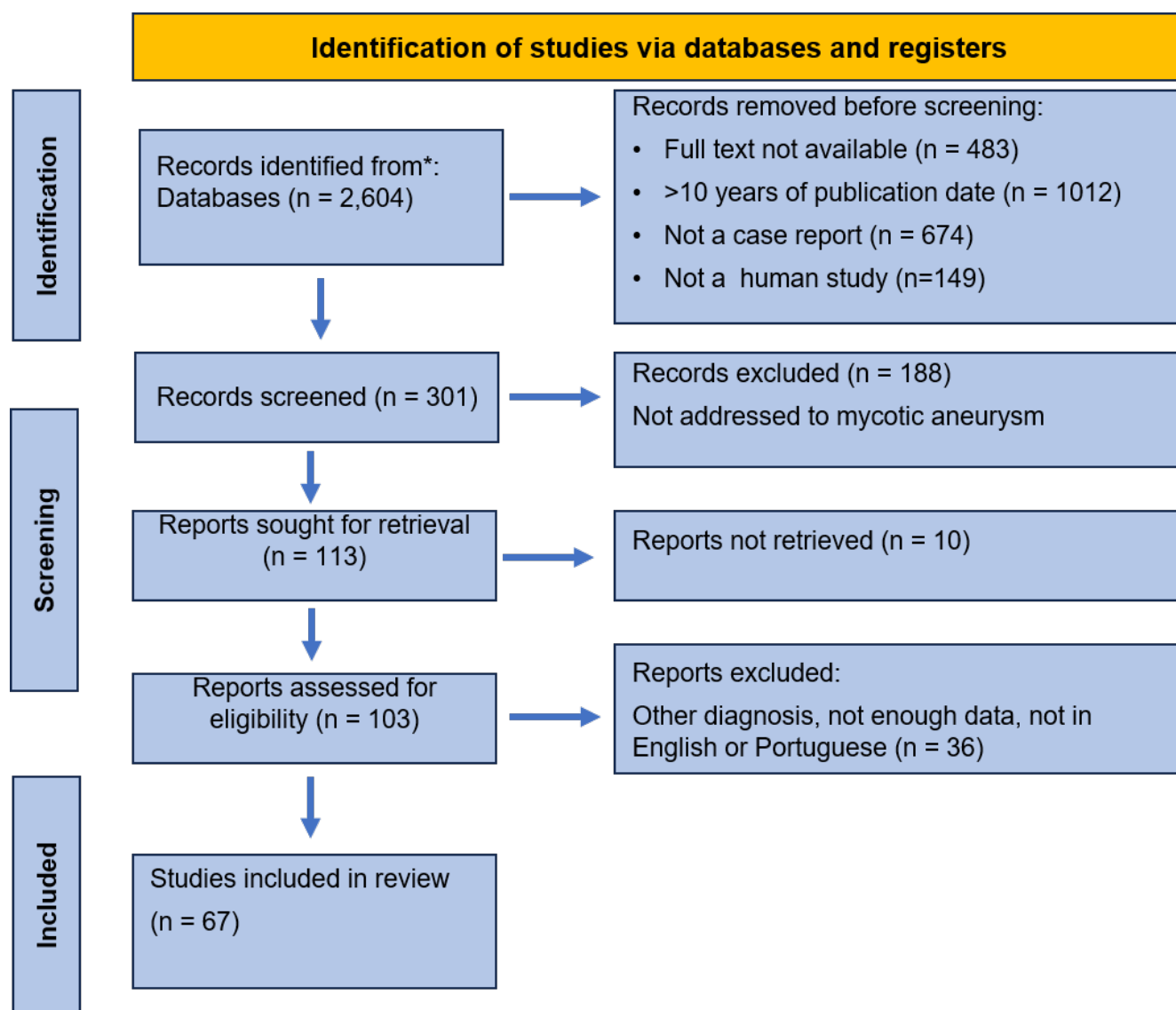


Figure 1. Literature search according to PRISMA guidelines.

Table 1. Summary of literature review.

Authors	Age/Sex	Initial Clinic Presentation	Pathogen	Aneurysm Location (s)/ Multiple Aneurysms?	Aneurysm Size (mm)	Aneurysm Rupture?	Treatment	Treatment Results	Associated antibiotic therapy	Outcomes
Gupta et al., 2013 ²	11 y/o; Female	Right-sided frontal headache, bilateral chemosis and eye-lid swelling, right-sided retro-orbital pain, ophthalmoplegia and ptosis. Right third, fourth and sixth nerve palsy, with reduced visual acuity more in the right eye 6/48 than the left 6/18	---	Intracavernous carotid	16.5×15.5×14.3 mm	No	Endovascular treatment with covered stent placement	---	Yes	Partial remission of symptoms
Flor-de-Lima et al., 2013 ⁴	17 y/o; Female	Headache, vomiting, drowsiness pupil asymmetry and cardiac systolic seagull-like murmur	Abiotrophia defectiva	M2 segment of right MCA	18×13 mm maximum length	Yes	Decompressive craniectomy Aneurysm clipping	No need for reoperation	Yes	Partial remission of symptoms
Abecassis et al., 2013 ¹⁵	35 y/o; Male	Intermittent visual disturbances in right eye preceding severe headaches with right homonymous hemianopsia	Streptococcus mitis, viridans and gordonii, plus a fungal infection	Left PCA at the P2-P3 junction	4 mm	Yes	Excision of the aneurysm with a right-sided parasagittal interhemispheric approach	Mild enlargement in an irregularity in the left PCA proximal to the occlusion that receded after 2 weeks of antifungal treatment	Yes	Total remission of symptoms
Ito et al., 2014 ¹⁶	74 y/o; Female	Found unconscious. Slightly disturbed consciousness but no localized neurological deficits. Developed generalized convulsions on day 30 of hospitalization	Streptococcus mutans	Left parietal artery	4 mm	Yes	Excision of the aneurysm with STA-MCA bypass	No need for reoperation	Yes	Total remission of symptoms
Saito et al., 2014 ¹⁷	78 y/o; Female	Sudden-onset left hemiparesis with dominance in the upper extremity	---	Right MCA (at the bifurcation)	---	Yes	No surgical intervention	---	Yes	The patient died as a result of the symptoms
Chinparlee et al., 2014 ¹⁸	16 y/o; Female	Severe headache, positive meningeal irritation signs without focal neurological deficit	Streptococcus viridans	Basilar artery	1.8 cm	Yes	Coil Embolization	No need for reoperation	Yes	Total remission of symptoms

MCA = middle cerebral artery; STA = superficial temporal artery; PCA = posterior cerebral artery; ACA = anterior cerebral artery; SMC = superior middle cerebral artery; PICA = posterior inferior cerebellar artery; ICICA = intracavernous internal carotid artery; SCA = superior cerebellar artery; PCoA = posterior communicating artery; DSA = digital subtraction angiography.

Table 1. Continued...

Authors	Age/Sex	Initial Clinic Presentation	Pathogen	Aneurysm Location (s)/ Multiple Aneurysms?	Aneurysm Size (mm)	Aneurysm Rupture?	Treatment	Treatment Results	Associated antibiotic therapy	Outcomes
Crowe et al., 2014 ¹⁹	48 y/o; Male	Fever, myalgia, night sweats and weight loss	Rothia aeria	Multiple Parieto-occipital branch of the right PCA (1) Pericallosal branch of the left ACA (2)	3-mm (1 and 2)	Yes	No surgical intervention	---	Yes	Total remission of symptoms
Lotan et al., 2014 ²⁰	23 y/o; Female	Behavioral changes, apathy, low grade fever, headache and fatigue. Mild left facial paralysis, mild left hemiparesis, left upper quadrantsia and fever	Enterococcus faecalis	Multiple Right MCA (M2 Sylvian segment) (1) Distal MCA bilaterally (2)	3 mm (1)	Yes	Endovascular embolization with platinum coils	No need for reoperation	Yes	Total remission of symptoms
Nelson et al., 2014 ²¹	41 y/o; Female	Worsening headache and vomiting. Left face, hand and foot numbness. Diminished light touch and temperature sensation in the region of the left cranial trigeminal nerve, mandibular division, left hand and the dorsal aspect of left foot, as well as 4/5 strength on left foot dorsiflexion	---	Right superior cerebellar artery	4.1 mm	---	No surgical intervention	---	Yes	Total remission of symptoms

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Table 1. Continued...

Authors	Age/Sex	Initial Clinic Presentation	Pathogen	Aneurysm Location (s)/ Multiple Aneurysms?	Aneurysm Size (mm)	Aneurysm Rupture?	Treatment	Treatment Results	Associated antibiotic therapy	Outcomes
Ding et al., 2014 ²²	35 y/o; old	Complained of intermittent diplopia	Streptococcus mitis	Multiple Ruptured aneurysm of the right distal pericallosal artery (1) Irregular left MCA M3 segment opercular aneurysm (2). Fusiform right MCA M4 segment posterior cortical branch aneurysm (3)	3 mm (1); 1.5 x 3 mm (2); 4x1 mm (3)	Yes	Endovascular stent-assisted coil embolization	No need for reoperation	Yes	Total remission of symptoms
Hill et al., 2014 ²³	64 y/o; Male	Increased somnolence and dysarthria	Pseudomonas aeruginosa	Right occipital cortical vessel	---	Yes	No surgical intervention	---	No	The patient died as a result of the symptoms
Urakami et al., 2014 ²⁴	74 y/o; Male	Aphasia and apraxia	Enterococcus faecium	Left temporo-frontal area	2 mm	Yes	No surgical intervention	---	Yes	Total remission of symptoms
Wang et al., 2015 ²⁵	22 y/o; Male	Vomiting and progressively deteriorated consciousness GCS was 5 with a fixed dilated right pupil and constricted left pupil	---	Multiple Distal Branch of the right PCA (1) Distal branch of the left MCA (2)	---	Yes	Embolization of the ruptured aneurysm with Onyx	No need for reoperation	Yes	Total remission of symptoms
Koffie et al., 2015 ²⁶	49 y/o; Male	Fevers, altered mental status, and generalized body aches	---	Right MCA	5 mm	Yes	Microsurgical clipping	No need for reoperation	Yes	The patient died as a result of the symptoms
Aoyama et al., 2015 ²⁷	66 y/o; Female	Dyspnea of sudden onset, and gradual reduction in the level of consciousness	Streptococcus agalactiae	---	---	Yes	---	---	Yes	The patient died as a result of the symptoms

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Table 1. Continued...

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Lee et al., 2015 ²⁸	46 y/o; Male	Right third, fourth, and sixth cranial nerve palsies; manifesting with complete ptosis, mydriasis, and total gaze paresis in the right eye	---	Right ICA	---	No	Endovascular stent	No need for reoperation	Yes	Partial remission of symptoms
Shinya et al., 2015 ²⁹	77 y/o; Male	Impaired consciousness (GCS of E1V1M5), right conjugate deviation of the eyes, and hemiparesis of the left upper and lower limbs	Aspergillus	ICA (1) PCA (P1-P2) (2)	10 mm, irregular (1)	Yes	Trapping of ICA with a STA bypass (1) Trapping of PCA (2)	No need for reoperation	Yes	The patient died as a result of the symptoms
Nonaka et al., 2016 ³⁰	Case 1: 24 y/o; Male Case 2: 47 y/o; Male	Case 1: 1-month history of headache and low-grade fever Case 2: cerebral infarction in the territory of the left PCA, which presented with a homonymous hemianopia	Case 1: Blood culture: Streptococcus sanguis Case 2: methicillin-resistant Staphylococcus aureus	Case 1: Distally saccular aneurysm lying on the angular branch of the left MCA Case 2: Nonruptured bilobular aneurysm at the left P3-P4 junction	Case 1: 4.5 x 4.0 mm saccular aneurysm lying Case 2: 10.0 x 8.0 mm	Case 1: Yes Case 2: No	Case 1 and 2: Endovascular therapy	Case 1 and 2: No need for reoperation	Case 1 and 2: Yes	Case 1 and 2: Total remission of symptoms
Maruyama et al., 2016 ³¹	26 y/o; Male	No significant neurologic deficits	Haemophilus parainfluenzae	Multiple	---	Yes	Microsurgical clipping	No need for reoperation	Yes	Total remission of symptoms
Fusco et al., 2016 ³²	Case 1: 24 y/o; Female Case 2: 33 y/o; Female	Case 1 and 2: No neurological deficits (IIA was an incidental finding after the diagnosis of ischemic stroke, in the postoperative period of valve replacement)	Case 1: Streptococcus mitis Case 2: ---	Case 1: Left distal MCA branch Case 2: Left M3 segment of MCA	Case 1: 5.3 mm x 4.7 mm Case 2: 4 mm	Case 1 and 2: No	Case 1 and 2: Coil embolization	Case 1 and 2: No need for reoperation	Case 1 and 2: Yes	Case 1 and 2: Total remission of symptoms
Schneider et al., 2016 ³³	42 y/o; Male	Change in mental status, unresponsive	Streptococcus viridans and Streptococcus gordonii	Multiple Right MCA (1) Right PCA (2) Left MCA (3)	3.9 mm x 2.7 mm x 2.3 mm (1) PCA = 5.1 mm x 5.3 mm x 6 mm (2)	Yes	Craniotomy and the insertion of a ventriculostomy tube	No need for reoperation	Yes	Partial remission of symptoms

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Table 1. Continued...

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Farran and Antony, 2016 ³⁴	60 y/o; Male	Headaches, fatigue, memory loss, and behavioral abnormalities	Nocardia abscessos	ICA	---	No	Drainage of the abscess with subsequent resection of the infected aneurysm	No need for reoperation	Yes	Total remission of symptoms
Muraoka et al., 2016 ³⁵	56 y/o; Male	Headache, loss of consciousness	Aspergillus	Right P2 segment of PCA	---	Yes	Endovascular	No need for reoperation	Yes	No remission of symptoms
Rhodes et al., 2016 ³⁶	Case 1: 50 y/o; Male	---	Case 1: Abiotrophia defectiva Case	Case 1: Distal M2 segment of Left MCA	Case 1: 5x3 mm Case 2: 4x3mm Case 3: --- Case 4: 3.8mm Case 5: 6 - 8 mm Case 6: ---	Cases 1, 2, 3 and 6: Yes Cases 4 and 5: No	Cases 1, 3 and 6: No surgical intervention Case 2: Aneurysmal resection, microvascular by-pass Cases 4 and 5: Microsurgical clipping	No need for reoperation	All cases: Yes	Cases 1, 2 and 3: Total remission of symptoms Cases 5 and 6: Partial remission of symptoms Case 4: The patient died as a result of the symptoms
	Case 2: 25 y/o; Female		2: Abiotrophia defectiva Case	Case 2: Left MCA Case 3: Right MCA (thrombosed)						
	Case 3: 48 y/o; Male		3: Granulicatella adiacens Case	MCA Case 3: Right MCA						
	Case 4: 60 y/o; Male		4: Abiotrophia defectiva Case	Case 4: Ruptured distal branch of right MCA Case 5: Multiple: Right and Left MCA and Left ACA						
	Case 5: 31 y/o; Male		5: Granulicatella adiacens Case	Case 6: Left frontal-parietal area (ruptured)						
	Case 6: 30 y/o; Male		6: Granulicatella adiacens	Petrous portion of the right ICA						
Han et al., 2016 ⁴	21 y/o; Male	Headache and vomiting; suddenly developed right facial palsy; severe neck stiffness	Klebsiella pneumoniae	Petrous portion of the right ICA	8 mm to 15mm	---	Endovascular balloon-expandable covered stent	No need for reoperation	Yes	Partial remission of symptoms
Morotti et al., 2016 ³⁷	Case 1: 35 y/o; Male	Case 1: Thunderclap headache after a 3-day fever Case	Case 1: Enterococcus Faecalis Case 2: ---	Case 1: Multiple: M4 division of MCA (unruptured) and high pericallosal aneurysm (ruptured) Case 2: Right calcarine artery	Case 1: --- Case 2: ---	Case 1 and 2: Yes	Case 1 and 2: Endovascular glue embolization	No need for reoperation	Case 1 and 2: Yes	Case 1 and 2: Total remission of symptoms
	Case 2: 60 y/o; Male	2: No neurological findings (IIA was an incidental finding after endocarditis)								
Khatibi et al., 2016 ³⁸	---	Severe headache and photophobia	Staphylococcus aureus	P2 segment of the right PCA	7x4 mm	Yes	Coil Embolization	No filling of the aneurysm	Yes	Total remission of symptoms

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Table 1. Continued...

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Champeaux et al., 2017 ²⁹	19 y/o; Male	Sudden loss of consciousness, at hospital (GCS 8)	Streptococcus mitis	Multiple Left distal PCA (junction P2-P3) and left distal PCA (junction P2-P3) (ruptured)	5 mm	Yes	Coil Embolization	No need for reoperation	Yes	Partial remission of symptoms
Piccirilli et al., 2017 ³¹	67 y/o; Male	Unresolved, severe and refractory to analgesic therapy neck pain from nine months. Mild tetraparesis, right hyper-reflexia and paresthesia	Group B Streptococcus	Fusiform aneurysm on the anterolateral medullary segment of the left PCA (unruptured)	15 X 12 mm in diameter, with a wide neck	Yes	Microsurgical clipping	No need for reoperation	Yes	Total remission of symptoms
Lin et al., 2017 ³²	25 y/o; Female	Severe headache and generalized seizure. GCS: 15. Left hemiparesis likely due to hemorrhage	Alpha-streptococcus	Distal right M2 aneurysm on MCA	8mm x 6mm x 6mm	Yes	Microsurgical clipping	Need for reoperation with coil and glue embolization due to acute bleeding around the right M2 aneurysm clips with recurrence and the development of a new left pericallosal aneurysm	Yes	Partial remission of symptoms
Shi et al., 2017 ³³	63 y/o; Male	Right blepharoptosis	---	Right ICICA	8.5 x 4.5 mm	No	Stent plus Embolization	No need for reoperation	Yes	Partial remission of symptoms
Kim et al., 2017 ³⁴	57 y/o; Female	Generalized weakness, generalized seizure with decreased in mental status	---	---	---	Yes	External ventricular drain (EVD)	---	Yes	Total remission of symptoms
Wang et al., 2017 ³⁵	25 y/o; Male	Sudden onset of dysarthria left hemiparesis, and generalized seizure. Initial NIH was 3	Gram-positive streptococci - Viridans	Origin of the M2 branche	5 mm x 3 mm	Yes	EVD and Coil Embolization	No need for reoperation	Yes	Partial remission of symptoms

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Table 1. Continued...

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Ohnake et al., 2017 ⁴⁴	Case 1: 80 y/o; Male Case 2: 36 y/o; Male with Marfan syndrome	Case 1: respiratory dysfunction and low-grade fever Case 2: high-grade fever, headache, deterioration of consciousness (GCS E1V1M2), and left hemiplegia	Case 1: Enterococcus faecium Case 2: blood culture test was negative	Case 1: Ipsilateral distal MCA aneurysm (ruptured) Case 2: Multiple. Distal left MCA aneurysm. After 3 new aneurysms in the anterior cerebral circulation	Case 1: --- Case 2: One: from 1.2 mm to 2.5 mm); the other 3: ---	Cases 1 and 2: Yes	Cases 1 and 2: Microsurgical clipping	No need for reoperation	Cases 1 and 2: Yes	Case 1: Total remission of symptoms Case 2: The patient died as a result of aspiration pneumonia
Glenn et al., 2017 ⁴⁵	51 y/o; Male	Right-sided facial droop, right upper extremity and right lower extremity weakness. GCS: 12, NIH: 22	---	Multiple Far lateral left SCA (1) Left M2-M3 branch junction (2)	---	Yes	---	---	Yes	The patient died as a result of the symptoms
Németh et al., 2017 ⁴⁶	68 y/o; Female	Severe otitis externa (otalgia and otorrhea) and bleeding on the contralateral (left) ear, left-sided mastoidectomy	---	Multiple Right ICA (1) Right common and internal carotid artery (2) Petrous segment of the left ICA (3)	---	---	Flow diverter stent	No need for reoperation	Yes	---
Garg and Khosroshahi, 2017 ⁴⁷	55 y/o; Male	Became delirious and was found to have an intracranial hemorrhage	Bartonella henselae	Multiple Distal M3 branch of the MCA (1) M2 branch of the MCA (2)	M3 = 4.1 x 4.4 x 5.1 x 1.3 mm M2 = 7.2 x 6.9 x 13.3 x 4.6 mm	Yes	---	---	Yes	Partial remission of symptoms
Nosse et al., 2018 ⁴⁸	17 y/o; Female	---	---	Left M4 segment of the MCA	8 mm	No	Microsurgical trapping	No need for reoperation	Yes	Total remission of symptoms
Ko et al., 2018 ⁸	14 y/o; Female	Nausea, vomiting, diarrhea, petechiae on both hands, and confused verbal orientation. Severe headache followed by loss of consciousness	Methicillin-sensitive Staphylococcus aureus (MSSA)	Multiple Left PCoA (ruptured) (1) Left-sided PCA aneurysm (2)	10mm (2)	Yes	Coil embolization	No need for reoperation	Yes	The patient died as a result of the symptoms

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Table 1. Continued...

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Liu et al., 2018 ¹⁹	28 y/o; Male	Severe headache, associated with nausea, vomiting and aphasia	---	M2 bifurcation of the left MCA	6 mm	Yes	Microsurgical clipping	No need for reoperation	Yes	Total remission of symptoms
Kobets et al., 2018 ²⁰	41 y/o; Female	Complete loss of vision in the left eye, left-leg weakness, and progressive lethargy. Bilateral proptosis with nonreactive pupils, complete bilateral third nerve palsy, and decreased left V1 and V2 sensation	Aspergillus fumigatus	Right cavernous ICA aneurysm	14.34 × 13.92 with a 5.3 mm neck	No	Pipeline embolization	No need for reoperation	Yes	Partial remission of symptoms
Boissonneau et al., 2018 ²¹	82 y/o; Male	Fall; Complete right visual loss	Campylobacter fetus	A1-A2 right junction of the ACA	25 x 20 mm	No	Microsurgical clipping	No need for reoperation	Yes	Partial remission of symptoms
Voruganti et al., 2018 ²²	56 y/o; Female	Fever, chills, severe headache, blurred vision, and change in mental status	Oxacillin resistant S. epidermidis	Multiple MCA (1) PCA (2)	---	Yes	Emergent left craniotomy with hematoma evacuation	---	Yes	The patient died as a result of the symptoms
Buchanan et al., 2019 ²³	26 y/o; Male	Persistent fevers, headache, nausea, and emesis. Overwhelming sudden-onset headache	Disseminated coccidioidomycosis	Multiple Bifurcation of the M2 division of the left MCA (1). Distally in an M3 segment (2) Saccular left A1 aneurysm and fusiform dilatations of both left M2 segments (3) 5 additional aneurysms in the contralateral ACA and MCA divisions (4)	---	Yes	Microsurgical clipping	No need for reoperation	Yes	Partial remission of symptoms

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Rangwala et al., 2019 ⁵⁴	27 y/o; Female	Severe headaches and blurred vision	---	Distal MCA	---	Yes	STA-MCA bypass and excision of distal MCA aneurysm	No need for reoperation	Yes	Partial remission of symptoms The patient died from another intraparenchymal hemorrhage not associated with vascular lesions
Daneshmand et al., 2019 ⁵⁵	30 y/o; Male	Recurrent face and arm tingling	Cardiobacterium hominis	Multiple A1: Frontal branch of the right MCA / A2: ---	A1: 4 mm A2: ---	No	A1: --- A2: Endovascular approach	No need for reoperation	Yes	Partial remission of symptoms
Boukobra et al., 2019 ⁵⁶	Case 1: 54 y/o; Male Case 2: 42 y/o; Male	Case 1: 10-day history of headache, fever and diffuse joint pain, II/VI systolic heart murmur and a II/VI Aortic insufficiency Case 2: fever, nausea and vomiting, worsening headache. Petechial bleeding on the trunk, face, and subungual hemorrhages, suggesting a diagnosis of IE An episode of partial seizures with secondarily generalized status epilepticus	Case 1: Streptococcus bovis Case 2: Methicillin-sensitive Staphylococcus aureus MSSA	Case 1: Saccular aneurysm at the bifurcation of a distal branch of the right PCA Case 2: Fusiform aneurysm of a distal branch of the left PCA	Case 1: 3 mm Case 2: ---	Case 1 and 2: No	Case 1 and 2: Endovascular embolization	Case 1 and 2: No need for reoperation	Case 1 and 2: Yes	Case 1 and 2: Total remission of symptoms
OuYang et al., 2019 ⁵⁷	49 y/o; Male	Headache, dizziness, altered mental status	---	Left internal carotid cavernous sinus	---	No	Stent-assisted coils	After 2 months post-surgery: DSA showed a recurrent aneurysm, but the patient declined reoperation	Yes	Total remission of symptoms The patient died from epistaxis

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Table 1. Continued...

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Gupta et al., 2019 ⁵⁸	5 y/o	Left-sided neck swelling	---	Proximal left ICA	4 x 6 x 5.5 cm	No	Coil embolization	No need for reoperation	Yes	Total remission of symptoms
Imamura et al., 2019 ⁵⁹	35 y/o; Male	High fever, left retro-orbital pain, ptosis, headache, diplopia; complete third, fourth, and sixth cranial nerve palsies on the left side	Brucella sp.	Multiple	---	No	Pipeline Embolization	No need for reoperation	Yes	Total remission of symptoms
Vieira et al., 2019 ⁶⁰	42 y/o; Female	Sudden-onset headache associated with vomiting, and significant neck stiffness (Hunt and Hess grade II)	Taenia solium - Neurocysticercosis	Frontal M2 segment of the left MCA associated with multiple areas of irregular caliber that corresponded to an arterial spasm	---	Yes	Direct clipping was not feasible for the dilatation, leading they to opt for wrapping the lesion	No need for reoperation	No	Total remission of symptoms
Hall et al., 2019 ⁷	8 y/o; Male	High-grade fever, altered mental status, tachycardia, poor perfusion	Streptococcus pneumoniae	Multiple 2 aneurysms in the left MCA branches arising from the left sylvian branches	Smaller one (3.4 mm); larger one (9.8 mm)	No	Coil embolization	No need for reoperation	Yes	Total remission of symptoms
Jain et al., 2019 ⁶¹	24 y/o; Female	Unilateral headache, retroorbital pain and progressive unilateral vision impairment. Decreased extraocular movements of the right eye and decreased visual acuity on the right to light perception only	Aspergillus	Fusiform pseudoaneurysm involving the cavernous segment of the right ICA	---	Yes	Coil Embolization	CT after 24 hours showed HSA and further extension of the aneurysm distally	No	The patient died as a result of the symptoms

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Ando et al., 2019 ¹⁰	Case 2: 43 y/o; Male Case 3: 51 y/o; Male	Case 2: sudden headache and sensory aphasia. Mild consciousness disturbance Case 3: sudden onset of headache and right hemianopia. Sudden headache with transient consciousness disturbance	Case 2: Enterococcus faecalis Case 3: Streptococcus salivarius	Case 2: partially thrombosed aneurysm at the proximal portion of MCA Case 3: fusiform aneurysm at the distal MCA. Subsequently, multiple aneurysms of the left distal PCA	Case 2: 50 mm Case 3: fusiform aneurysm: 3.5 mm	Case 2: No Case 3: Yes	Case 2: Endovascular ICA occlusion and revascularization Case 3: Coil Embolization of the fusiform aneurysm. Subsequently, endovascular treatment using internal trapping with coils of the multiple aneurysms	Case 2 and 3: No need for reoperation	Case 2: No Case 3: Yes	Case 2: Total remission of symptoms Case 3: Partial remission of symptoms
Alshari et al., 2019 ¹²	35 y/o; Male	After a dental procedure, he developed numbness affecting his left upper limb, then progressing to affect both legs. He was found collapsed. Neck stiffness and agitation	Streptococcus agalactiae, sensitive to penicillin	Multiple fusiform aneurysms involving two distal branches of the right MCA	---	Yes	No surgical intervention	---	Yes	Total remission of symptoms
Shashidhar et al., 2020 ¹³	32 y/o; Male	Fever, headache, vomiting, and seizures for a week. Signs of meningeal irritation	Alpha hemolytic streptococci	Basilar artery	---	Yes	No surgical intervention	---	Yes	The patient died as a result of the symptoms
Beckerman et al., 2020 ¹⁴	Case 1: 21 y/o; Male Case 2: 20 y/o; Male	Cases 1 and 2: No neurological deficits - preoperative finding	Case 1: Bartonella henselae Case 2: Bartonella vinsonii	Case 1: Right MCA (M3) Case 2: Left frontal MCA	Case 1: 2.5x2mm Case 2: 1.3 cm x 1 cm	Cases 1 and 2: No	Cases 1: Microsurgical clipping Case 2: Resection and Ligation	Case 1 and 2: No need for reoperation	Yes	Total remission of symptoms
Wang et al., 2020 ¹⁵	53 y/o; Male	Sudden severe headache and loss of consciousness, dilation of left pupil and disappearance of light reaction	---	Basilar Artery (original sites of bilateral SCA and PCA were also involved)	---	Yes	No surgical intervention	---	Yes	The patient died as a result of the symptoms
Shiba et al., 2020 ¹⁶	52 y/o; Male	---	Genella morbillorum	M4 segment of the right MCA	6.9 x 4.0 x 5.2 mm	No	Microsurgical trapping	No need for reoperation	Yes	Total remission of symptoms

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Savić et al., 2020 ⁶⁵	13 y/o; Female	Sudden loss of consciousness, right-side weakness. GCS: 7. Motor power of the right limbs was 0 out of 5.	---	Frontoparietal branch of the left MCA in the M2 segment	8 x 7 mm, with a base of 2.3 mm	Yes	Microsurgical trapping and EVD	Three days after surgery: left-sided brain edema, the level of consciousness dropped to GCS: 8	No	No remission of symptoms
Kalousek et al., 2020 ⁶⁷	61 y/o; Male	Occipital headache	---	Multiple Three in the M4 segments of both MCAs	1.85 x 2.76 mm 1.73 mm 1.81 mm	Yes	Coil embolization only for the ruptured aneurysm	No need for reoperation	No	Total remission of symptoms
Fisher et al., 2021 ⁶⁸	50 y/o; Male	Acutely developed right hemiparesis and aphasia	---	Aneurysm in the M2 segment of the left MCA (ruptured)	---	Yes	No surgical intervention	---	Yes	The patient died as a result of the symptoms
Kisilevsky et al., 2021 ⁶⁹	62 y/o; Male	Intermittent aphasia, left-sided temporal headache and eye pain	Rothia dentocariosa	Fusiform aneurysm of a distal branch of the left MCA	---	Yes	Endovascular embolisation	No need for reoperation	Yes	Partial remission of symptoms
Samples et al., 2021 ⁷⁰	10 y/o; Female	7 days of headaches, lethargy, and fever	Staphylococcus aureus and Streptococcus anginosus	2 aneurysms involving the left M1 segment of MCA	1.6 x 0.9 mm 4.05 x 4.28	Yes	Coil embolization only for the ruptured aneurysm	No need for reoperation	Yes	Partial remission of symptoms
Mian et al., 2021 ⁷¹	48 y/o; Male	Dysarthria with left-sided weakness; ill-appearing drowsy with mild left-sided weakness. Acute aphasia	Aspergillus fumigatus	PCA	2mm	Yes	No surgical intervention	---	Yes	The patient died as a result of the symptoms
Somboonithiphol et al., 2021 ⁷³	8 month -old; Female infant	Prolonged high-grade fever; suddenly developed seizures and drowsiness; normotonia with equal movement of all extremities, and positive Brudzinski and Kernig signs	Pseudomonas aeruginosa	Lobulated fusiform aneurysm at the proximal basilar artery	8 mm in maximal diameter and 13 mm in length	Yes	Ventriculostomy to relieve hydrocephalus. Partial coiling of the aneurysm and coiling of the left of the left vertebralbasilar junction	No need for reoperation	Yes	Total remission of symptoms
Nemtergian et al., 2021 ⁷²	54 y/o; Female	Acute intense headaches, dysphasia, and epileptic seizures	---	M3 segment of the MCA	---	Yes	Microsurgical clipping	No need for reoperation	No	Total remission of symptoms

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Mitsui et al., 2021 ⁷³	55 y/o; Male	Visual field defect	Streptococcus oralis	Multiple Left angular artery (1) Right MCA (2)	---	Yes	Endovascular embolization	No need for reoperation	Yes	---
Matsuzono et al., 2021 ⁷⁴	63 y/o; Female	Aphasia and dysarthria	Staphylococcus haemolyticus	Left MCA (M2/M3)	2 mm	Yes	Endovascular embolization	No need for reoperation	Yes	Total remission of symptoms

MCA = middle cerebral artery; STA = superficial temporal artery; PCA = posterior cerebral artery; ACA = anterior cerebral artery; SMC = internal carotid artery; PICA = posterior inferior cerebellar artery; IICA = intracavernous internal carotid artery; SCA = superior cerebellar artery; PCoA = posterior communicating artery; DSA = digital subtraction angiography.

headache, nausea, and monoparesis of the right lower limb. Neurological examination revealed a Glasgow Coma Scale (GCS) score of 14, postural instability, nuchal rigidity (positive Brudzinski sign), cranial tenderness, grade IV strength in the upper limbs and the right lower limb, and grade III strength in the left lower limb.

A cranial CT scan showed hypoattenuating extra-axial collections in the left frontal region with a maximum thickness of 0.9 cm and adjacent to the left interhemispheric falx with a maximum thickness of 1.2 cm, exhibiting pachymeningeal enhancement, sulcal and gyral effacement on that side, and compression of the ipsilateral lateral ventricle. Soft tissue content was observed in the left maxillary and frontal sinuses, suggestive of acute sinusitis.

Based on these findings, the diagnostic hypothesis of complicated sinusitis with cerebral empyema/bacterial meningoencephalitis was proposed. The patient developed septic shock and secondary intracranial hypertension. He underwent surgery for cerebral empyema drainage/left frontoparietal decompressive craniectomy without complications. Two days later, the otolaryngology team performed drainage of the left maxillary, ethmoidal, and frontal sinuses. The patient stabilized, showed clinical improvement, and was discharged one week after the surgical procedures.

The patient returned to the neurosurgery clinic without complaints. Neurological examination revealed left-sided facial asymmetry, motor deficits in the left upper limb and right lower limb, and right-sided asymmetric hemiparesis with grade IV strength in the left upper limb and grade III strength in the right lower limb, along with a positive Babinski sign on the right.

A follow-up cranial CT scan revealed ventricular asymmetry with dilation of the left temporal and occipital horns, a subdural collection with liquor-like density in the left frontotemporal and parietal regions, and gliosis in the left frontal region, without signs of active infection. The team opted to monitor the patient, with the potential need for a ventriculoperitoneal shunt before cranioplasty, depending on the persistence of asymmetry, ventricular dilation, or signs of intracranial hypertension.

Six months after the initial presentation, the patient returned following head trauma (a punch) at the craniectomy site, presenting with sweating, perioral and upper extremity pallor,

a syncopal episode, and local pain. Neurological examination showed bulging in the left temporal region without tenderness, GCS 15, asymmetric right hemiparesis with worse proximal weakness (grade III in the right upper limb, grade IV in the right lower limb), and left-sided facial asymmetry.

A CT scan revealed a large left temporal intraparenchymal hemorrhage, cerebral edema, and ventricular dilation. The patient underwent surgery for hemorrhage drainage and placement of a high-pressure ventriculoperitoneal shunt. Postoperative CT showed left temporal gliosis, no evidence of bleeding, and reduced ventricular size.

Twelve days postoperatively, the patient experienced altered consciousness, significant edema at the craniectomy site, worsening right upper limb strength (grade II), and spasticity. A new cranial CT scan indicated rebleeding at the previous hematoma site, with moderate volume, mass effect, herniation at the decompressive craniotomy site, and midline shift. MRI showed a large clot, precluding exclusion of arteriovenous malformation or other vascular disease. Angiography identified a 3-mm aneurysm in the M3-M4 segment of the middle cerebral artery, with a suspected mycotic aneurysm (Figure 2). CSF cultures, blood cultures, and urinalysis showed no specific pathogens. Echocardiogram and transesophageal echocardiogram were normal, without evidence of cardiac or infectious abnormalities.

Due to the aneurysm's location, surgical challenges, and anatomical variations, endovascular treatment was chosen. The patient was referred to the neurosurgery team at another tertiary hospital, with an endovascular team. A week later, he returned, reporting spontaneous aneurysm thrombosis, confirmed by cranial CT showing complete hematoma resolution.

The patient remained stable, without motor deficit worsening, and was discharged with instructions for outpatient follow-up. Cranioplasty was performed three months later without complications. At follow-up a week after cranioplasty and three months post-aneurysm thrombosis, the patient was hemodynamically stable and asymptomatic. Neurological examination showed GCS 15, right-sided paresis with spasticity in the upper limb (grade IV strength in the right lower limb, grade V in other limbs), mild left-sided facial asymmetry, and gradual improvement of deficits.

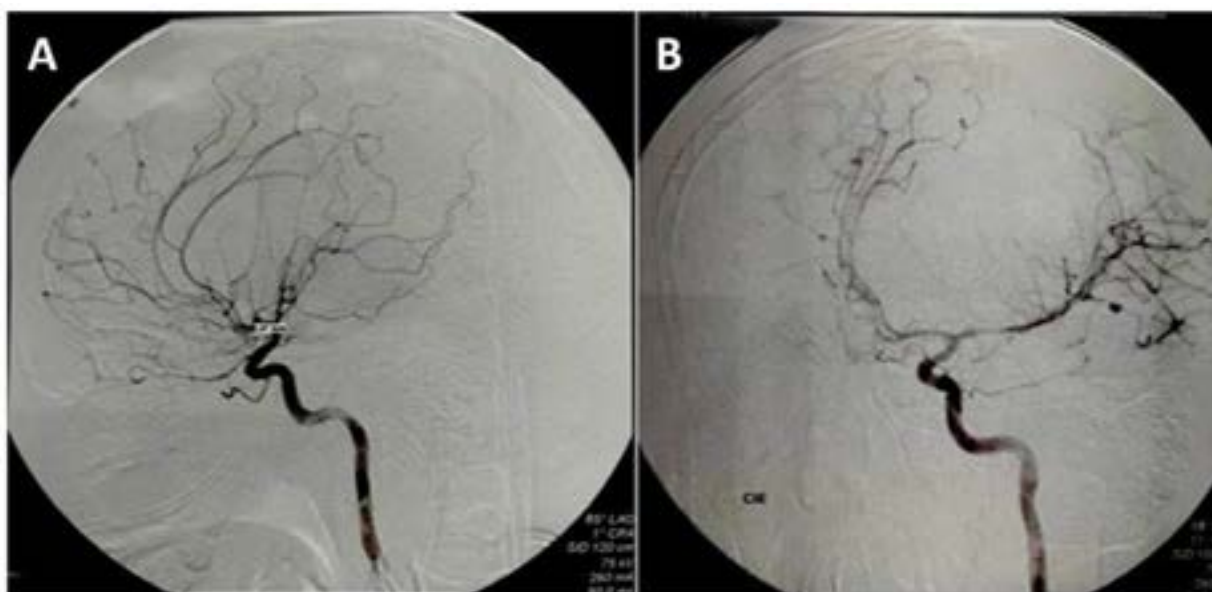


Figure 2. A and B. Cerebral angiography images showing aneurysmal dilation in the M3-M4 segment of the left Middle Cerebral Artery (MCA), approximately 3 mm in size.

RESULTS

Demographic profile

The predominant sex was male, constituting 66.23% of the sample. Regarding age and gender distribution among patients with mycotic aneurysms, certain demographic groups showed a predominance. The most affected age groups were 48-55 years (19.48%), followed by 24-31 years (16.88%) and 56-63 years (12.98%). The average age of the cases analyzed was 41 years.

Initial neurological presentation

The analysis of the initial clinical presentation revealed that the most common neurological symptoms were headache (37%), visual and ocular disturbances (27.16%), such as changes in visual acuity and visual fields, and retro-orbital pain; neuromuscular disorders (22.22%), such as weakness, hemiparesis, and ptosis; altered level of consciousness (13.58%); seizures (9.87%); language and speech disorders (9.87%), such as aphasia and dysarthria; and, less prominently, sensory disturbances (6.17%).

Etiological agent

In 27.2% of the evaluated cases, no specific etiological agent was identified. Among the rest, 86.44% presented bacterial agents, while the remainder had fungal (11.86%) and helminthic agents,

showing a frequency of bacteria 6.5 times higher. Among the bacterial agents, the *Streptococcus* genus was predominant, accounting for 34.37% of all pathogens identified.

Characteristics of the aneurysms

The location, laterality, size, and number of aneurysms were analyzed. The most common location was the Middle Cerebral Artery (53.3%), followed by the Posterior Cerebral Artery (17.7%) and the Internal Carotid Artery in its intracranial portion (12.2%). Involvement of other structures, such as the Basilar, Anterior Cerebral, Pericallosal, and Superior Cerebellar arteries, accounted for 12.2% of cases. Other structures accounted for 4.4% of cases, including the Calcarine, Posterior Communicating, Anterior Communicating, and Posterior Inferior Cerebellar arteries. In terms of laterality, there was a slight predominance of aneurysms on the left side, constituting 53.4% of the cases. The largest aneurysm diameter, after its maximum growth, ranged from 1.3 mm to 50 mm, with an average of 7.08 mm, a standard deviation of 7.4 mm, and a median of 4.5 mm. The presence of single aneurysms was observed in a significant proportion, constituting 64.5% compared to multiple aneurysms. Additionally, 67.4% of cases had an episode of intracranial hemorrhage.

Treatment

Among 80 patients evaluated regarding surgical interventions, 77.5% underwent some procedure for aneurysm correction (excluding ventriculoperitoneal shunts, decompressive

craniotomies, etc.). Of the 62 patients who underwent surgery, 58% underwent endovascular intervention, while the remaining patients underwent microsurgery. Antibiotic therapy was prescribed for 74 of the evaluated patients (91.3%). Of these, 75.6% underwent associated surgical procedures, while in the remaining cases, antibiotics were the only therapy used.

Clinical outcome

Among the cases analyzed for remission of neurological symptoms, 51.4% experienced complete remission and remained without deficits after treatment, whether clinical or surgical. Additionally, 27.1% showed partial improvement, while 21.4% did not show any improvement in the clinical neurological condition. Death associated with the aneurysm occurred in 20% of cases, with 81.25% of these patients showing no remission of symptoms after treatment.

DISCUSSION

AIIs are pseudoaneurysms characterized by an inflammatory process that extends beyond the adventitia into the muscular layer, potentially associated with septic emboli^{3,9,39}. These formations lack a true wall and are instead characterized by a fragile single layer of connective tissue that usually impedes distal blood flow. Approximately 2 to 5% of neurological complications in individuals with infective endocarditis manifest as AIIs, but the actual incidence remains unknown due to limited brain screening in patients without neurological symptoms^{6,12}. It is interesting to note that our patient did not show any significant cardiac alterations, given the normal results of their electrocardiogram and transesophageal echocardiogram, contrasting with the typical pattern of AIIs occurring secondarily to infective endocarditis.

Two predominant mechanisms contribute to the development of AIIs: intra-arterial dissemination, in which septic emboli travel to distal cerebral vessels, often originating from an infected heart valve due to infective endocarditis; and extra-arterial dissemination, where vascular infection arises from infected neighboring structures, as seen in cases associated with cavernous thrombophlebitis, meningitis, sinus infections, extension from an infected focus, hematogenous seeding of the intima during bacteremia, or trauma^{3,4}. The underlying pathophysiological processes of intra-arterial dissemination involve the vasa

vasorum hypothesis, in which the infection progresses from the inside out of the arterial wall; or the embolic hypothesis, characterized by the centrifugal progression of the intima into the adventitia^{5,75}. In our case, we present a patient who developed an AII approximately one year after an episode of bacterial cerebral empyema. After discussion, the team concluded a probable extra-arterial development of the AII caused by the extension of the infectious focus associated with trauma suffered by the patient.

In the pediatric population, 57% to 91% of AIIs are caused by *Streptococcus viridans* and *Staphylococcus aureus*^{3,12,39}. *Pneumococcus* is considered a rare etiological agent in children, although it is increasingly identified, especially in children with underlying congenital heart disease or previous endocarditis⁷. In the present study, the most common pathogens were *Staphylococcus aureus* and *Aspergillus*. Pathogens such as *Streptococcus viridans*, *Staphylococcus aureus*, *Streptococcus anginosus*, *Abiotrophia defectiva*, *Streptococcus pneumoniae*, and *Pseudomonas aeruginosa* were also identified in pediatric cases. Despite the low incidence of fungal infections in our study (11.86%), 33.3% of the deaths with a defined etiological agent were associated with *Aspergillus*, despite occurring 6.5 times less than bacterial agents in the total cases. Thus, although less frequent, fungal infections appear to have higher mortality.

Diagnostic criteria for AIIs include typical imaging findings, identification of predisposing infections, and consideration of clinical indicators, with early diagnosis being crucial for reducing morbidity and mortality^{12,75}. However, this identification is difficult as patients may be asymptomatic or present with subacute and nonspecific symptoms, such as fever, headache, nausea, vomiting, chills, malaise, and mild focal deficits⁷. In pediatric patients, these symptoms may be even more underreported. In this review, 78% of reported cases presented some neurological alteration in their initial clinical presentation, with most being nonspecific, predominated by headache and changes in acuity and visual field. Symptoms such as fever, nausea, and vomiting were also predominant, accounting for 36% of cases. Our patient also presented a nonspecific picture, with the rebleeding of their previous lesion leading to a neurovascular study that assessed the presence of the aneurysm.

The evaluation of the cases in this systematic review allowed for the construction of a profile with the highest incidence of infectious aneurysms in terms of gender and age group. Regarding the more affected gender, the male population presented a higher

prevalence of cases (66.23%). Similar values were observed for mortality rates, with 62.5% of the 16 deaths being male. Regarding age group, the most affected group was those between 48 and 55 years old (19.48%). Regarding mortality rates, this group accounted for 37.5% of the deaths studied. The analysis of these data reinforces a typical profile of higher incidence risk, with the male population aged 48 to 55 years^{8,12,13}. However, only the gender aligns with our patient's profile, as, due to their age of 11 years, they belong to the pediatric population (0 to 21 years), an age group that accounted for only 16.88% of cases in this study. In this population, AIIs are rare, comprising 15% of all pediatric intracranial aneurysms, and are underdiagnosed due to their low rupture rate (2%)^{1,3}.

Cerebral angiography is the gold standard for diagnosing AIIs, with common characteristics including distal location, fusiform shape, poorly defined aneurysm neck, irregular contours, and multiplicity^{7,8,12,37,75}. Regarding location, infectious aneurysms are preferentially found in the arteries of the anterior circulation (79.2%), especially in the middle cerebral artery, affected in 56.9% of cases^{3,7,13,75}. AIIs in the posterior circulation are observed in 20.8% of cases^{8,12,39}.

The cases evaluated in the present study reinforce the prevalence of aneurysms in the anterior circulation and the preference for branches of the MCA, which were affected in 53.3% of the total aneurysms. The occurrence of posterior circulation aneurysms also aligned with the frequency described by Alawieh et al., as they were identified in 26.8% of our cases¹³. Moreover, posterior circulation aneurysms were present in 57.1% of patients who died, while this location corresponds to only 26.8% of the total aneurysms evaluated. These data may indicate a worse prognosis for posterior circulation AIIs compared to anterior circulation AIIs.

IIAs tend to be small, with multiple occurrences reported in 19.4% of cases^{12,13,75}. In the present study, the average size of the largest dimensions of aneurysms was 7.08 mm, with aneurysms identified ranging from 1.3 mm to 50 mm. The occurrence of multiple AIIs was identified in 35.5%, slightly higher than the frequency found by Alawieh et al.¹³. The dimensions of our patient's AII are consistent with the literature, as it was a single aneurysm measuring about 3 mm, affecting distal branches of the MCA.

Although there is no established standard treatment for infectious aneurysms, the use of specific antibiotics is one of the cornerstones

of therapy^{3,12,40,75}. Among the total cases analyzed in the present study, 20% died, while mortality among cases that did not receive antibiotics was 50%. Additionally, a relationship was observed between the absence of antibiotic therapy and the persistence of initial neurological symptoms. Of the total patients evaluated, the persistence or worsening of neurological symptoms was observed in 21.4% of cases, while among patients who did not use antibiotics, this statistic was 40%. These data highlight the importance of combining antibiotics in the treatment of AIIs. Our patient adhered to antibiotic therapy and showed partial remission of symptoms after treatment.

Surgical approaches are especially recommended in cases of hemorrhagic events and progressive aneurysm enlargement, given the high morbidity and mortality rates associated with these conditions^{3,6,40,75}. While microsurgical techniques include aneurysm resection and clipping, endovascular approaches involve embolization using coils, liquid agents, and flow-diverting stents^{3,8,12,13}. While 22.5% of the total cases in this study did not undergo surgical intervention, 62.5% of patients who died did not have surgery, reinforcing the importance of these interventions in the prognosis of AIIs. Furthermore, the type of surgery performed did not show a clear relationship with mortality, as among the 6 patients who died and underwent surgery, 50% had endovascular correction, while 50% had microsurgical correction. In our patient's case, AII correction surgery was not performed because it was identified during endovascular intervention that the aneurysm had undergone spontaneous thrombosis.

The average mortality rate for AIIs analyzed in the systematic review by Alawieh et al.¹³ is 24.1%, which can reach 80% when evaluating ruptured AIIs¹³. In the present study, the overall mortality rate for evaluated AIIs is 20%, while mortality for AIIs with ICH or in ruptured aneurysms was 25.4%.

CONCLUSION

Intracranial infectious aneurysms are rare complications, particularly in the pediatric population. This study reports a case of a mycotic aneurysm secondary to a brain abscess in a pediatric patient who was no longer in an infectious process. The wide range of clinical findings highlights the importance of imaging exams for early diagnosis and the definition of

appropriate neurosurgical management. The high mortality rate justifies the screening of infectious foci (especially for Infective Endocarditis) and underscores the importance of considering them as a differential diagnosis in the presence of spontaneous intracranial hemorrhage.

Despite the relevance of the findings, this review has some limitations. The included evidence is mostly based on case reports and small case series, which may introduce publication bias and limit the generalizability of the results. Additionally, the review process was constrained by the lack of standardized outcome reporting across studies, and no formal assessment of risk of bias or certainty of evidence was performed. Nevertheless, the synthesis of available data offers important insights for clinical practice. Early recognition of intracranial infectious aneurysms—especially in the context of unexplained hemorrhagic events—may guide timely investigation of infectious sources and influence surgical decision-making. For future research, multicenter registries and prospective studies are needed to improve the understanding of optimal management strategies, particularly in pediatric patients.

REFERENCES

- Eddleman CS, Surdell D, DiPatri A Jr, Tomita T, Shaibani A. Infectious intracranial aneurysms in the pediatric population: endovascular treatment with Onyx. *Childs Nerv Syst.* 2008;24(8):909-15. <http://doi.org/10.1007/s00381-008-0614-8>. PMID:18365209.
- Gupta V, Jain V, Mathuria S, Khandelwal N. Endovascular treatment of a mycotic intracavernous carotid artery aneurysm using a stent graft. *Interv Neuroradiol.* 2013;19(3):313-9. <http://doi.org/10.1177/159101991301900308>. PMID:24070080.
- Somboonnithiphon K, Chanthanaphak E, Ayudhaya SPSN, Khongkhatithum C, Sirilert B. Successful endovascular treatment of pediatric basilar infectious (mycotic) aneurysm: a case report and review of the literature. *Childs Nerv Syst.* 2021;37(8):2687-93. <http://doi.org/10.1007/s00381-020-04917-8>. PMID:33030602.
- Han MS, Jung SH, Kim TS, Joo SP. Reconstructive endovascular treatment of an intracranial infectious aneurysm in bacterial meningitis: a case report and review of literature. *World Neurosurg.* 2016;90:700.e1-5. <http://doi.org/10.1016/j.wneu.2016.02.031>. PMID:26893038.
- Wang X, Chen G, Li M, et al. Rapid formation and rupture of an infectious basilar artery aneurysm from meningitis following suprasellar region meningioma removal: a case report. *BMC Neurol.* 2020;20(1):94. <http://doi.org/10.1186/s12883-020-01673-9>. PMID:32171270.
- Hamisch CA, Mpotsaris A, Timmer M, et al. Interdisciplinary treatment of intracranial infectious aneurysms. *Cerebrovasc Dis.* 2016;42(5-6):493-505. <http://doi.org/10.1159/000448406>. PMID:27598469.
- Hall JM, McElroy BJ, Arora PK, Mohandas S. Intracranial mycotic aneurysm complicating *Streptococcus pneumoniae* infection. *J Pediatr.* 2019;211:223.e1. <http://doi.org/10.1016/j.jpeds.2019.03.031>. PMID:31005282.
- Piccirilli M, Prizio E, Cannizzaro D, Tropeano MP, Guidetti G, Santoro A. The only case of mycotic aneurysm of the PICA: clinical-radiological remarks and review of literature. *J Clin Neurosci.* 2017;38:62-6. <http://doi.org/10.1016/j.jocn.2016.12.034>. PMID:28118952.
- Ko H, Kim G, Lee HD, Choi KH, Sung SC. Ruptured intracranial aneurysm in an adolescent with infective endocarditis. *Pediatr Int.* 2018;60(4):376-7. <http://doi.org/10.1111/ped.13513>. PMID:29573363.
- Ando K, Hasegawa H, Kikuchi B, et al. Treatment strategies for infectious intracranial aneurysms: report of three cases and review of the literature. *Neurol Med Chir.* 2019;59(9):344-50. <http://doi.org/10.2176/nmc.oa.2019-0051>. PMID:31270285.
- Akimoto K, Yanaka K, Nakamura K, et al. Simultaneous intracerebral and subarachnoid hemorrhages caused by multiple infectious intracranial aneurysms treated endovascularly and by microsurgical clipping: illustrative case. *J Neurosurg Case Lessons.* 2022;3(7). <http://doi.org/10.3171/CASE21685>. PMID:36130552.
- Ducruet AF, Hickman ZL, Zacharia BE, et al. Intracranial infectious aneurysms: a comprehensive review. *Neurosurg Rev.* 2009;33(1):37-46. <http://doi.org/10.1007/s10143-009-0233-1>. PMID:19838745.
- Alawieh A, Chaudry MI, Turner RD, Turk AS, Spiotta AM. Infectious intracranial aneurysms: a systematic review of epidemiology, management, and outcomes. *J Neurointerv Surg.* 2018;10(7):708-16. <http://doi.org/10.1136/neurintsurg-2017-013603>. PMID:29463620.
- Flor-de-Lima F, Lisboa L, Sarmiento A, Almeida J, Mota T. Mycotic brain aneurysm and cerebral hemorrhagic stroke: a pediatric case report. *Eur J Pediatr.* 2013;172(9):1285-6. <http://doi.org/10.1007/s00431-013-2032-5>. PMID:23686512.
- Abecassis IJ, Adel JG, Ayer A, Batjer HH. A ruptured infectious intracranial aneurysm with a combined fungal and bacterial etiology. *Clin Neurol Neurosurg.* 2013;115(11):2393-6. <http://doi.org/10.1016/j.clineuro.2013.08.026>. PMID:24034820.
- Ito H, Tanaka Y, Sase T, et al. Cerebral hyperperfusion syndrome following the excision of a mycotic aneurysm with superficial temporal artery-to-middle cerebral artery bypass: case report. *Neurol Med Chir.* 2014;54(10):845-50. <http://doi.org/10.2176/nmc.cr2013-0017>. PMID:24257489.
- Saito A, Kawaguchi T, Hori E, et al. Subarachnoid Hemorrhage After an Ischemic Attack Due to a Bacterial Middle Cerebral Artery Dissecting Aneurysm: Case Report and Literature Review. *Neurol Med Chir.* 2014;54(3):196-200. <http://doi.org/10.2176/nmc.cr2012-0251>. PMID:24140774.

18. Chimparlee N, Jittapiromsak P, Tantivatana J, Chattranukulchai P. Classical complication of infective endocarditis: ruptured, large mycotic cerebral aneurysm. *BMJ Case Rep.* 2014;2014:bcr2013202275. <http://doi.org/10.1136/bcr-2013-202275>. PMID:24686799.
19. Crowe A, Ding NS, Yong E, Sheorey H, Waters MJ, Daffy J. Rothia aerea mitral valve endocarditis complicated by multiple mycotic aneurysms: laboratory identification expedited using MALDI-TOF MS. *Infection.* 2013;42(2):419-23. <http://doi.org/10.1007/s15010-013-0532-x>. PMID:24078192.
20. Lotan E, Orion D, Bakon M, Kuperstein R, Greenberg G. Ruptured intracranial mycotic aneurysm in infective endocarditis: radiological and clinical findings. *Isr Med Assoc J.* 2014;16(5):317-9. PMID:24979841.
21. Nelson G, Fermo OP, Thakur KT, et al. Resolution of a fungal mycotic aneurysm after a contaminated steroid injection: a case report. *BMC Res Notes.* 2014;7(1):327. <http://doi.org/10.1186/1756-0500-7-327>. PMID:24885172.
22. Ding D, Raper DM, Carswell AJ, Liu KC. Endovascular stenting for treatment of mycotic intracranial aneurysms. *J Clin Neurosci.* 2014;21(7):1163-8. <http://doi.org/10.1016/j.jocn.2013.11.013>. PMID:24518267.
23. Hill JA, Mokadam NA, Rakita RM. Intracranial mycotic aneurysm associated with left ventricular assist device infection. *Ann Thorac Surg.* 2014;98(3):1088-9. <http://doi.org/10.1016/j.athoracsur.2013.10.094>. PMID:25193193.
24. Urakami T, Hamada Y, Magarihuchi H, Yamakuchi H, Aoki Y. Enterococcal endocarditis complicated with ruptured infected-intracranial aneurysm: with pharmacokinetic-pharmacodynamic documentation in proof of the successful antimicrobial treatment. *J Infect Chemother.* 2014;20(12):810-3. <http://doi.org/10.1016/j.jiac.2014.07.011>. PMID:25153621.
25. Wang K, Sun J, Zhang X, Zhang Q, Chen Z. Management of consecutive development of ruptured intracranial mycotic aneurysms: case report. *Turk Neurosurg.* 2015;25(2):310-2. <http://doi.org/10.5137/1019-5149.jtn.6814-12.1>. PMID:26014019.
26. Koffie RM, Stapleton CJ, Torok CM, Yoo AJ, Leslie-Mazwi TM, Codd PJ. Rapid growth of an infectious intracranial aneurysm with catastrophic intracranial hemorrhage. *J Clin Neurosci.* 2015;22(3):603-5. <http://doi.org/10.1016/j.jocn.2014.09.007>. PMID:25455738.
27. Aoyama R, Kobayashi A, Tubokou Y, et al. Two case reports of group B streptococcal infective endocarditis complicated by embolism. *Intern Med.* 2015;54(18):2333-6. <http://doi.org/10.2169/internalmedicine.54.4709>. PMID:26370857.
28. Lee B, Kim C, Carrasco J. Intracranial infectious aneurysm in orbital cellulitis. *Orbit.* 2015;34(4):175-8. <http://doi.org/10.3109/01676830.2015.1014515>. PMID:25955309.
29. Shinya Y, Miyawaki S, Nakatomi H, et al. Recurrent cerebral aneurysm formation and rupture within a short period due to invasive aspergillosis of the nasal sinus; pathological analysis of the catastrophic clinical course. *Int J Clin Exp Pathol.* 2015;8(10):13510-22. PMID:26722566.
30. Nonaka S, Oishi H, Tsutsumi S, et al. Endovascular Therapy for Infectious Intracranial Aneurysm: A Report of Four Cases. *J Stroke Cerebrovasc Dis.* 2016;25(3):e33-7. <http://doi.org/10.1016/j.jstrokecerebrovasdis.2015.11.033>. PMID:26738813.
31. Maruyama R, Yamada A, Sugiyama T, et al. Mitral valve repair for endocarditis can be performed 3 days after repair of a bleeding mycotic brain aneurysm. *J Thorac Cardiovasc Surg.* 2016;151(4):e59-61. <http://doi.org/10.1016/j.jtcvs.2015.10.026>. PMID:26616464.
32. Fusco MR, Stapleton CJ, Griessenauer CJ, Thomas AJ, Ogilvy CS. Endovascular treatment of intracranial infectious aneurysms in eloquent cortex with super-selective provocative testing: case series and literature review. *Interv Neuroradiol.* 2016;22(2):148-52. <http://doi.org/10.1177/1591019915617326>. PMID:26672110.
33. Schneider MA, Pomidor MA. Cerebral mycotic aneurysm and infective endocarditis: a case study. *J Neurosci Nurs.* 2016;48(2):100-4. <http://doi.org/10.1097/JNN.0000000000000188>. PMID:26871240.
34. Farran Y, Antony S. Nocardia abscessus-related intracranial aneurysm of the internal carotid artery with associated brain abscess: a case report and review of the literature. *J Infect Public Health.* 2016;9(3):358-61. <http://doi.org/10.1016/j.jiph.2015.11.009>. PMID:26724261.
35. Muraoka S, Araki Y, Izumi T, Takeuchi K, Okamoto S, Wakabayashi T. Cerebral infarction and subarachnoid hemorrhage caused by central nervous system aspergillus infection. *World Neurosurg.* 2016;90:705.e9-13. <http://doi.org/10.1016/j.wneu.2016.03.021>. PMID:26996735.
36. Rhodes HM, Hirigoyen D, Shabnam L, Williams DN, Hansen GT. Infective endocarditis due to Abiotrophia defectiva and Granulicatella spp. complicated by infectious intracranial cerebral aneurysms: a report of three cases and review of the literature. *J Med Microbiol.* 2016;65(6):493-9. <http://doi.org/10.1099/jmm.0.000260>. PMID:27046228.
37. Morotti A, Gamba M, Costa P, et al. Infective endocarditis presenting with intracranial bleeding. *J Emerg Med.* 2016;51(1):50-4. <http://doi.org/10.1016/j.jemermed.2016.04.003>. PMID:27236244.
38. Khatibi K, Heit JJ, Telischak NA, Elbers J, Do HM. Republished: Cerebral vascular findings in PAPA syndrome: cerebral arterial vasculopathy or vasculitis and a posterior cerebral artery dissecting aneurysm. *J Neurointerv Surg.* 2016;8(8):e29. <http://doi.org/10.1136/neurintsurg-2015-011753.rep>. PMID:26122324.
39. Champeaux C, Walker N, Derwin J, Grivas A. Successful delayed coiling of a ruptured growing distal posterior cerebral artery mycotic aneurysm. *Neurochirurgie.* 2017;63(1):17-20. <http://doi.org/10.1016/j.neuchi.2016.10.005>. PMID:28283187.
40. Lin CT, Tranmer B, Durham S, Johnson D, Hamlin M, Bolman RM 3rd. Ruptured mycotic aneurysm and cerebral vasospasm in the setting of endocarditis and heart failure requiring cardiothoracic surgery: case report and literature review. *World Neurosurg.* 2017;100:711.e13-8. <http://doi.org/10.1016/j.wneu.2017.01.076>. PMID:28153625.
41. Shi L, Zhou M, Xu S, Wu Q, Yan W, Zhang J. Endovascular treatment of intracavernous internal carotid aneurysm secondary to pituitary infection. *World Neurosurg.* 2017;101:816.e5-9. <http://doi.org/10.1016/j.wneu.2017.02.076>. PMID:28238872.

42. Kim JS, Kang MK, Cho AJ, Seo YB, Kim KI. Complicated infective endocarditis: a case series. *J Med Case Rep.* 2017;11(1):128. <http://doi.org/10.1186/s13256-017-1274-7>. PMID:28482860.
43. Wang JL, Hinduja AP, Powers CJ. Successful coil embolization of a ruptured mycotic aneurysm that developed three days after septic embolic infarction: case report and review of the literature. *J Clin Neurosci.* 2017;39:95-8. <http://doi.org/10.1016/j.jocn.2017.01.021>. PMID:28209306.
44. Ohtake M, Tateishi K, Ikegaya N, Iwata J, Yamanaka S, Murata H. Initial treatment strategy for intracranial mycotic aneurysms: 2 case reports and literature review. *World Neurosurg.* 2017;106:1051.e9-16. <http://doi.org/10.1016/j.wneu.2017.07.016>. PMID:28711529.
45. Glenn J, Strecker-McGraw M, McGraw I, Jabbar K, James NA, Stone CK. Rupture of an occult intracranial mycotic aneurysm after intravenous thrombolysis with recombinant tissue plasminogen activator for acute ischemic stroke. *J Emerg Med.* 2017;53(5):717-21. <http://doi.org/10.1016/j.jemermed.2017.08.032>. PMID:28988732.
46. Németh T, Szakács L, Bella Z, Majoros V, Barzó P, Vörös E. The treatment of pseudoaneurysms with flow diverters after malignant otitis externa. *Interv Neuroradiol.* 2017;23(6):609-13. <http://doi.org/10.1177/1591019917729804>. PMID:28992722.
47. Garg S, Khosroshahi AA. 55-Year-Old male presenting with altered mental status: a complicated case of intracranial aneurysm. *Arthritis Care Res.* 2017;69(12):1915-9. <http://doi.org/10.1002/acr.23409>. PMID:28881419.
48. Nossek E, Setton A, Chalif DJ. Trapping and resection of cortical MCA mycotic aneurysm in eloquent area. *Acta Neurochir.* 2018;160(3):579-82. <http://doi.org/10.1007/s00701-017-3401-z>. PMID:29170845.
49. Liu W, Li C, Liu X, Xu Z, Kong L. Case of subarachnoid hemorrhage caused by tuberculous aneurysm. *World Neurosurg.* 2018;110:73-8. <http://doi.org/10.1016/j.wneu.2017.10.128>. PMID:29097336.
50. Kobets AJ, Scoco A, Nakhla J, et al. Flow-diverting stents for the obliteration of symptomatic, infectious cavernous carotid artery aneurysms. *Oper Neurosurg.* 2017;14(6):681-5. <http://doi.org/10.1093/ons/oxp166>. PMID:28961750.
51. Boissonneau S, Graillon T, Meyer M, Brunel H, Fuentes S, Dufour H. Intracranial giant mycotic aneurysm without endocarditis and vasculitis: report of rare entity and review of literature. *World Neurosurg.* 2018;119:353-7. <http://doi.org/10.1016/j.wneu.2018.08.086>. PMID:30144607.
52. Voruganti D, Gajurel K, Bhama JK, Cotarlan V. Ruptured intracranial mycotic aneurysm in infective endocarditis with left ventricular assist device and implantable cardiac defibrillator device: a clinical course. *Transplant Proc.* 2018;50(10):4064-6. <http://doi.org/10.1016/j.transproceed.2018.08.027>. PMID:30577316.
53. Buchanan IA, Ravina K, Strickland B, et al. Multiple intracranial aneurysms from coccidioidal meningitis: case report featuring aneurysm formation and spontaneous thrombosis with literature review. *World Neurosurg.* 2019;121:117-23. <http://doi.org/10.1016/j.wneu.2018.08.220>. PMID:30201579.
54. Rangwala SD, Strickland BA, Rennert RC, et al. Ruptured mycotic aneurysm of the distal circulation in a patient with mucormycosis without direct skull base extension: case report. *Oper Neurosurg.* 2019;16(3):E101-7. <http://doi.org/10.1093/ons/opy127>. PMID:29800469.
55. Daneshmand A, Rangel-Castilla L, Rydberg C, Wijdicks E. Ultra-rapid developing infectious aneurysms. *Neurocrit Care.* 2019;30(2):487-9. <http://doi.org/10.1007/s12028-018-0585-x>. PMID:30088210.
56. Boukobza M, Duval X, Laissy JP. Mycotic intracranial aneurysms rupture presenting as pure acute subdural hematoma in infectious endocarditis. Report of 2 cases and review of the literature. *J Clin Neurosci.* 2019;62:222-5. <http://doi.org/10.1016/j.jocn.2018.12.035>. PMID:30638783.
57. OuYang M, Huang X, Wang Y. Endovascular treatment of infectious pseudoaneurysm of internal carotid artery. *World Neurosurg.* 2019;125:42-3. <http://doi.org/10.1016/j.wneu.2019.01.147>. PMID:30731201.
58. Gupta R, Patro SK, Chauhan N, Kumar A. A giant pseudoaneurysm mimicking retropharyngeal abscess in a child. *Pediatr Emerg Care.* 2019;35(5):e79-83. <http://doi.org/10.1097/PEC.0000000000001100>. PMID:28328693.
59. Imamura H, Sakai N, Alexander MJ. Flow-diverter stenting of intracavernous internal carotid artery mycotic aneurysm. *J Stroke Cerebrovasc Dis.* 2019;28(7):e81-2. <http://doi.org/10.1016/j.jstrokecerebrovasdis.2019.04.026>. PMID:31101401.
60. Vieira E, Faquini IV, Silva JL, et al. Subarachnoid neurocysticercosis and an intracranial infectious aneurysm: case report. *Neurosurg Focus.* 2019;47(2):E16. <http://doi.org/10.3171/2019.5.FOCUS19280>. PMID:31370019.
61. Jain A, Goyal K, Meher R, Passey JC. Internal carotid artery bleed: a rare complication of invasive sphenoidal aspergillosis. *World Neurosurg.* 2019;129:292-4. <http://doi.org/10.1016/j.wneu.2019.06.014>. PMID:31203084.
62. Afshari FT, Al-Lawati K, Chavda S, Billing S, Flint G. Cerebral mycotic aneurysms secondary to Streptococcus Agalactiae induced infective endocarditis. *Br J Neurosurg.* 2019;33(6):693-5. <http://doi.org/10.1080/02688697.2017.1409873>. PMID:29183165.
63. Shashidhar A, Bharath RD, Satishchandra P, Rao MB, Arimappamagan A. Dissecting aneurysm of the basilar artery: a rare complication of bacterial meningitis in a postoperative case of CSF Rhinorrhea. *Neurol India.* 2020;68(1):173-5. <http://doi.org/10.4103/0028-3886.279691>. PMID:32129272.
64. Beckerman Z, Martínez-Bravo LE, Johnson G, Holt B, Fraser CD. Rare presentation of endocarditis and mycotic brain aneurysm. *Ann Thorac Surg.* 2020;109(3):e179-81. <http://doi.org/10.1016/j.athoracsur.2019.06.073>. PMID:31425670.
65. Shiba M, Toma N, Ikezawa M, et al. Intracranial-intracranial bypass using a Y-shaped artery graft for growing unruptured gemella morbillorum infectious aneurysm on artery supplying sensory cortex. *World Neurosurg.* 2020;142:328-33. <http://doi.org/10.1016/j.wneu.2020.07.056>. PMID:32683008.

66. Savić D, Alsheikh TM, Alhaj AK, et al. Ruptured cerebral pseudoaneurysm in an adolescent as an early onset of COVID-19 infection: case report. *Acta Neurochir.* 2020;162(11):2725-9. <http://doi.org/10.1007/s00701-020-04510-7>. PMID:32720013.
67. Kalousek V, Sajko T, Splavski B, et al. Current Endovascular Management of Infected Distal Branch Intracranial Aneurysms: a Case Report and Insight into the Literature. *Acta Clin Croat.* 2020;59(4):754-60. <http://doi.org/10.20471/acc.2020.59.04.24>. PMID:34285448.
68. Fisher B, Seese L, Mathier MA, Sultan I, Kilic A. Ruptured cerebral mycotic aneurysm in a left ventricular assist device patient with bacteremia. *Int J Artif Organs.* 2020;44(1):65-7. <http://doi.org/10.1177/0391398820925493>. PMID:32475219.
69. Kisilevsky E, Pesin N, Mandell D, Margolin EA. Rothia dentocariosa causing intracranial mycotic aneurysm and ischaemic stroke. *BMJ Case Rep.* 2021;14(3):e240349. <http://doi.org/10.1136/bcr-2020-240349>. PMID:33664036.
70. Samples DC, Ravindra VM, Thoms DJ, Tarasiewicz I, Grandhi R. Successful flow diversion treatment of ruptured infectious middle cerebral artery aneurysms with the use of Pipeline Flex with Shield technology. *Interv Neuroradiol.* 2021;27(2):225-9. <http://doi.org/10.1177/1591019921990506>. PMID:33509016.
71. Mian I, Ives S, Jean-Louis G, Lacznia A. Recurrent stroke and fatal ruptured mycotic aneurysm caused by invasive aspergillus fumigatus infection. *WMJ.* 2021;120(1):82-4. PMID:33974773.
72. Nerntengian N, Gkasdaris G, Baretas N, Theodoropoulou E, Birbilis T. The use of real-time 3D intraoperative ultrasound “angiography” in localization and occlusion control of a ruptured mycotic aneurysm: a case report. *J Neurol Surg A Cent Eur Neurosurg.* 2021;82(5):500-4. <http://doi.org/10.1055/s-0040-1720988>. PMID:33278825.
73. Mitsui K, Oda R, Lee T, et al. Multiple mycotic aneurysms with infective endocarditis: a case report. *J Infect Chemother.* 2021;27(10):1513-6. <http://doi.org/10.1016/j.jiac.2021.05.014>. PMID:34049794.
74. Matsuzono K, Ishiyama Y, Higaki A, et al. Successful endovascular coiling of infectious cerebral aneurysm following Staphylococcus haemolyticus endocarditis. *J Int Med Res.* 2021;49(11):3000605211058857. <http://doi.org/10.1177/03000605211058857>. PMID:34846922.
75. Esenkaya A, Duzgun F, Cinar C, et al. Endovascular treatment of intracranial infectious aneurysms. *Neuroradiology.* 2015;58(3):277-84. <http://doi.org/10.1007/s00234-015-1633-2>. PMID:26700825.

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Microsurgical Clipping of Bihemispheric Anterior Cerebral Aneurysm

Clipagem Microcirúrgica de Aneurisma Cerebral Anterior Bi-hemisférico

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Steven Awyono¹ 

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ABSTRACT

The bihemispheric anterior cerebral artery is a normal variant of the anterior cerebral artery. Aneurysm in the distal anterior cerebral artery with this variant presents a surgical challenge as parent artery occlusion must be avoided at any cost to prevent ischemia of the distal brain tissue. Several surgical procedures have been employed to treat distal anterior cerebral aneurysms. The pterional approach is considered a simple approach that can provide golden rules in aneurysm clipping even to the distal segment of the anterior cerebral artery. This report presents a rare case of an 83-year-old adult male with a bihemispheric anterior cerebral artery with aneurysm on the dominant side of the A2. We used the pterional approach that gives early proximal control and sufficient exposure to evaluate the parent artery. We successfully visualized the parent artery, neck of the aneurysm and the distal A2 segment. Successful clipping, achieved with patent efferent artery flow, was evaluated using transcranial doppler.

Keywords: Distal anterior cerebral artery; Pterional approach; Cerebral aneurysm

RESUMO

A artéria cerebral anterior bi-hemisférica é uma variante anatômica da artéria cerebral anterior. A presença de um aneurisma na artéria cerebral anterior distal com essa variante representa um desafio cirúrgico, pois a oclusão da artéria principal deve ser evitada a todo custo para prevenir isquemia do tecido cerebral distal. Diversas técnicas cirúrgicas têm sido empregadas no tratamento de aneurismas da artéria cerebral anterior distal. A abordagem pterional é considerada simples e eficaz, podendo fornecer os princípios fundamentais da clipagem de aneurismas, mesmo nos segmentos distais da artéria cerebral anterior. Este relato apresenta um caso raro de um paciente do sexo masculino, 83 anos, com artéria cerebral anterior bi-hemisférica e aneurisma no lado dominante do segmento A2. Utilizamos a abordagem pterional, que proporcionou controle proximal precoce e exposição adequada para avaliação da artéria principal. Conseguimos visualizar com sucesso a artéria principal, o colo do aneurisma e o segmento distal de A2. A clipagem foi realizada com êxito, mantendo o fluxo arterial aferente, avaliado por doppler transcraniano.

Palavras-Chave: Artéria cerebral anterior distal; Abordagem pterional; Aneurisma cerebral

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INTRODUCTION

The variability in the anterior cerebral artery (ACA) architecture is considerable. A wide range of ACA variations, including bihemispheric, azygous, crossover branches, and accessory ACA, have been documented in the literature¹⁻⁶. A notable surgical challenge arises when confronted with the distal ACA aneurysm accompanied by a bihemispheric ACA variant on the dominant side^{1-4,6}. When the fundus of an aneurysm extends medially or posteriorly, its visualization has the potential to be obstructed by the dominant artery. Here, we describe a case of an aneurysm in the right distal ACA, originating from the dominant side of a bihemispheric ACA variant. We describe its unique characteristics and surgical management using the pterional approach for this aneurysm.

CASE ILLUSTRATION

Examination

An 83-year-old man was brought to the emergency department with a loss of consciousness for seven hours prior. He had a history of uncontrolled hypertension for ten years. A neurological examination was obtained with the Glasgow coma scale of 4t and positive meningeal sign. Head computed tomography (CT) scan revealed acute extensive subarachnoid hemorrhage over the basal cistern expanding to both lateral cistern and anterior interhemispheric fissure with panventricular hemorrhage and ventricular enlargement (Figure 1A-B). Further CT angiography revealed a distal ACA aneurysm on the right side with a neck width of 6mm, dome width of 11 mm, and dome/neck ratio of 1.8 (Figure 1C-D). After discussing with the patient's family, we plan to perform aneurysm clipping and external ventricular drainage.

Operative technique

After induction of general anesthesia, the patient was positioned supine with gentle head extension and 45 degrees left rotation (Figure 1E). We used two layers of skin flap with right pterional craniotomy. After opening the bone flap, we visualized the subarachnoid hemorrhage over the cerebral convexity. Brain relaxation was achieved after performing cerebrospinal fluid drainage from the sylvian cistern. After adequate brain relaxation,

we visualized the distal internal cerebral artery (ICA) and followed it to expose the A1 segment of the right ACA (Figure 2A). In order to safely dissect the arachnoidal layer, proximal control was applied to the distal ICA. The brain spatula gently retracted the frontal lobe to enhance visualization of the ACA distally (Figure 2B). Following the surgical procedure involving the resection of the ipsilateral gyrus rectus, we successfully located and identified a fusiform aneurysm on the anterior communicating artery on its anterior side (Figure 2C). Bilateral A2 segment of the ACA was visualized, in which the left A2 Segment was hypoplastic. After adequate dissection of the arachnoidal layer, we could visualize the ipsilateral A2 segment (Figure 2D), which was then chosen as the proximal control to reduce blood flow through the aneurysm, hence mitigating the risk of intraoperative rupture. A slight head rotation towards the right side was performed to visualize the neck of the aneurysm, which projected in a posteromedial direction. A meticulous arachnoidal dissection technique was employed, utilizing a dissector instrument to expose the aneurysmal neck safely and carefully. Following a careful dissection procedure in the surroundings of the A2 segment of the right ACA, we successfully identified the efferent artery of the A2 on the ipsilateral side (Figure 3A).

We used a curved Yasargil titanium Clip® (blade length 6.8 mm) on the aneurysm (Figure 3B). Complete obliteration was achieved, and the ipsilateral blood flow of the A2 segment was confirmed using intraoperative doppler ultrasound. The blister aneurysm on the anterior communicating artery was then packed with muscle and fibrin glue to prevent rupture. The postoperative course was uneventful. On postoperative day 5, the patient depended on the ventricular drainage system, which then converted to a permanent ventriculoperitoneal shunt.

In this case, we performed the pterional approach to fulfill the golden rule of aneurysm clipping. These rules refer to the proximal and distal control with adequate exposure of the ACA branches⁷⁻¹⁰. This approach provides early identification of the proximal ACA, to apply the proximal control. Further arachnoidal dissection followed by resecting the ipsilateral rectus gyrus may enhance the visualization of the ACA segment. In using the pterional approach to treat distal ACA, we need to clearly expose the neck of the aneurysm to prevent incomplete clipping⁸.

Compared to the interhemispheric approach, the pterional approach is more widely accepted due to technical ease, familiarity, and early access of the proximal vascular to control

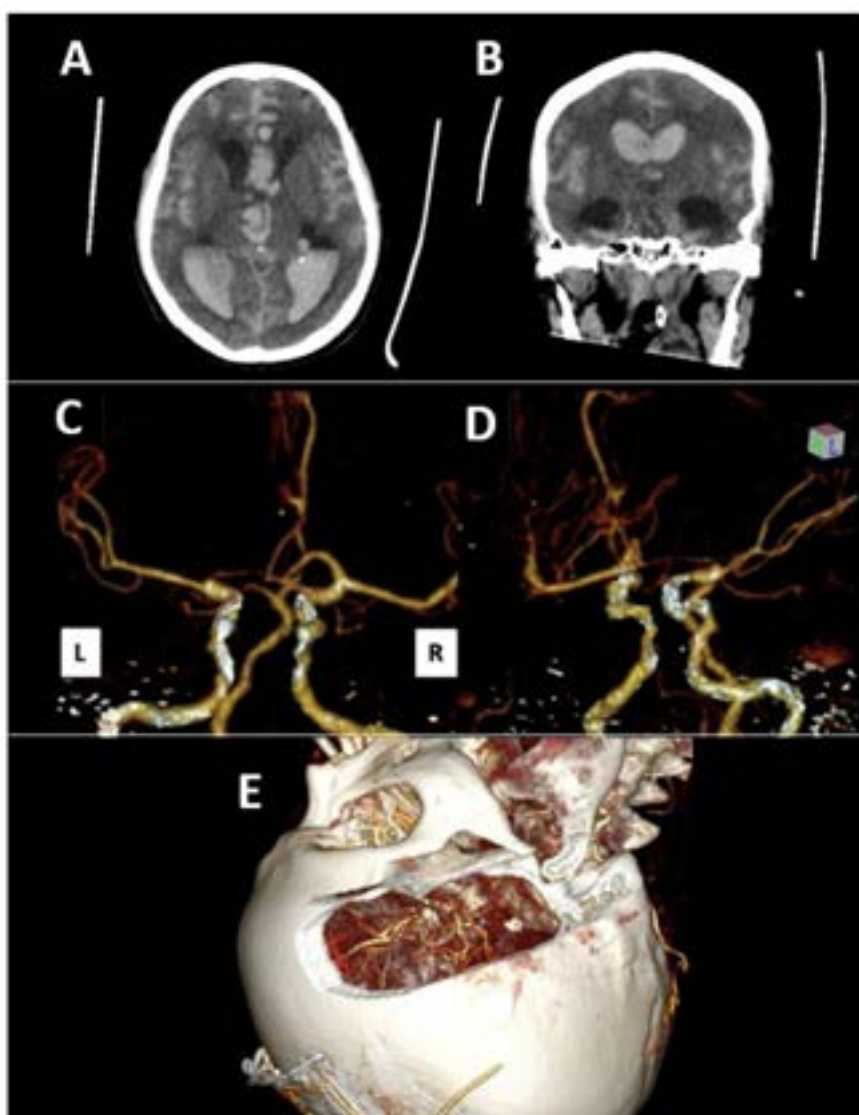


Figure 1. Preoperative head CT imaging showing the subarachnoid over the interhemispheric fissure with panventricular hemorrhage and ventricular enlargement **A.** Axial. **B.** Coronal. CT Angiography showing hypoplastic A2 on the left side with Saccular aneurysm on the left side of A2. **C.** Posterior view. **D.** 45° oblique view from left anterior side. **E.** Preoperative 3D planning to expose the aneurysm by using the pterional approach.

the A1 segment⁹. This approach also facilitates safe dissection of the arachnoidal trabeculae surrounding the aneurysm. It also evaluates the patency of parent artery to prevent the occlusion. However, severe brain retraction and narrow surgical exposure appear as disadvantages. Therefore, the resection of rectus gyrus is very helpful in enhancing the exposure. Compared to interhemispheric approach, the brain transgression, dissecting the olfactory tract and the division of superior sagittal sinus were not required. However, although the parasagittal bridging veins can be sacrificed, this can disturb the venous blood flow through frontal

ascending veins^{7,9-12}. In addition, the risk of venous infarction increases with brain retraction. The difficulty and laboriousness of dissecting the interhemispheric fissure should be considered due to be tightly opposed and adherence of the fissure. Lastly, early proximal control is not possible. On the contrary, the incidence of incomplete clipping was found to be higher in pterional approach cases (36%) than in the interhemispheric approach, especially when neck width is >8 mm. Therefore, to minimize this risk, safely dissecting and visualizing the aneurysm neck could help to properly and completely clip the aneurysm neck^{11,12}.

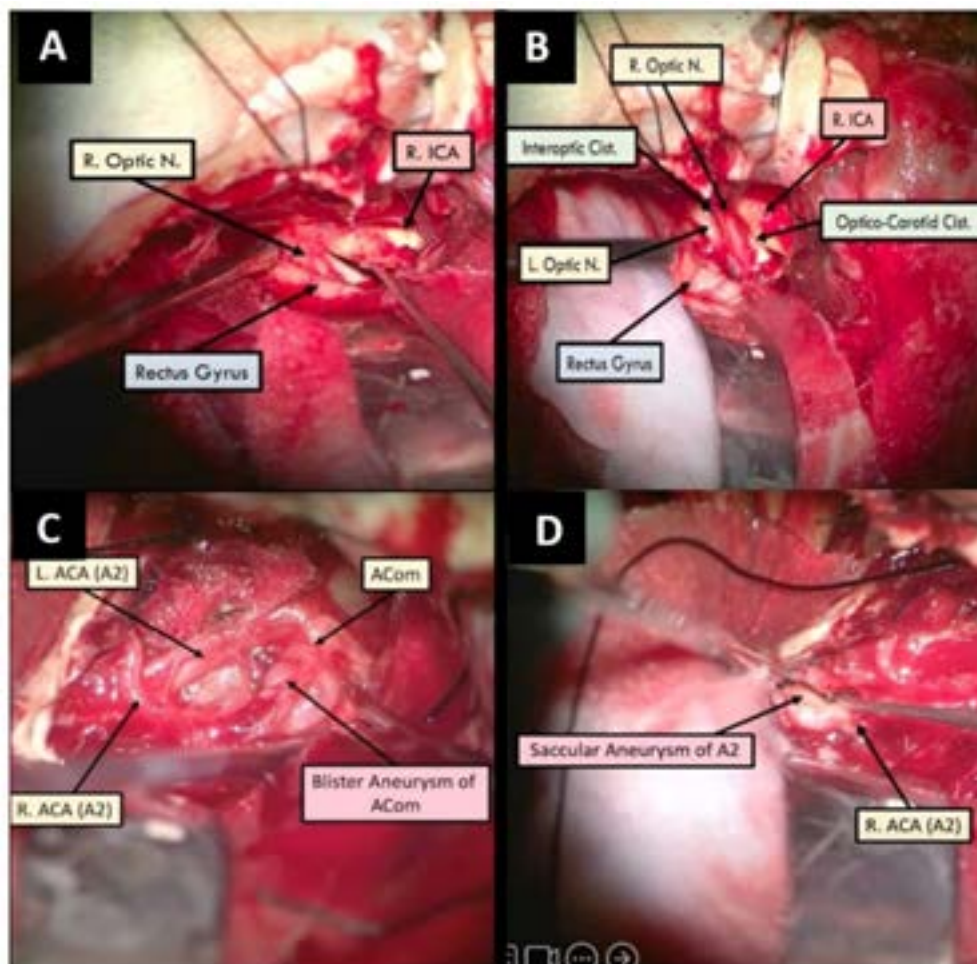


Figure 2. **A.** Initial dissection and visualization of the anatomical landmark. **B.** Further dissection exposing the optico-carotid cistern, bilateral optic nerve and part of the rectus gyrus to be resected. **C.** After resecting the ipsilateral rectus gyri, we can visualize the bilateral A1, Acom and bilateral A2 in which the left side A2 is hypoplastic. **D.** Further dissection of the right A2 to expose the aneurysm.

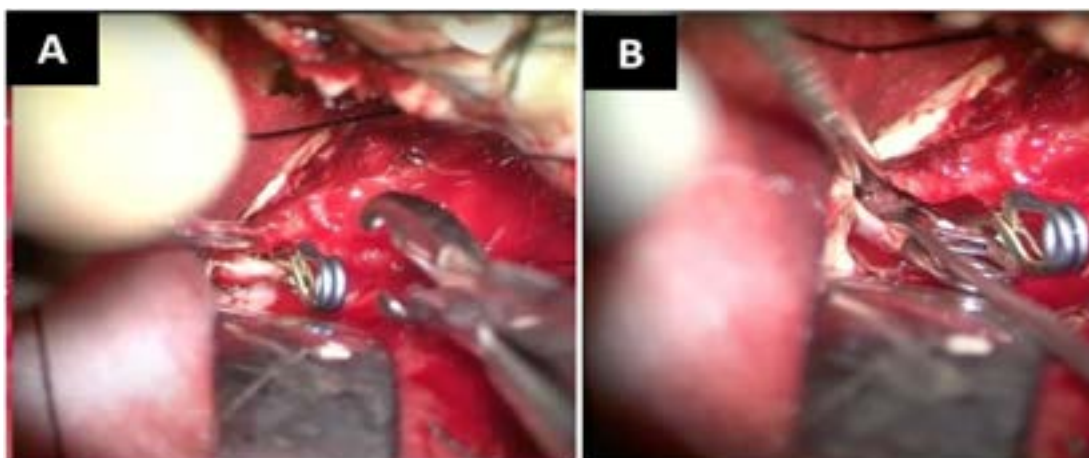


Figure 3. **A.** temporary clipping on the proximal A2 segment. **B.** Permanent clipping on the neck of the aneurysm using a curved titanium clip.

CONCLUSIONS

A preoperative evaluation is important to determine the anatomical variant of ACA aneurysm. There are two common approaches for ACA aneurysms which are the pterional and the interhemispheric. The pterional approach may be considered a safe and simple approach to treat distal ACA aneurysm while also fulfilling the surgical goals by giving wide exposure and early access to the proximal vessel.

REFERENCES

1. Lawton MT. Seven aneurysms: tenets and techniques for clipping. Stuttgart: Thieme Publishers Series; 2011. <http://doi.org/10.1055/b-002-66278>.
2. Cilliers K, Page BJ. Detailed description of the anterior cerebral artery anomalies observed in a cadaver population. *Ann Anat*. 2016;208:1-8. <http://doi.org/10.1016/j.aanat.2016.04.036>. PMID:27237980.
3. Tahir RA, Haider S, Kole M, Griffith B, Marin H. Anterior cerebral artery: variant anatomy and pathology. *J Vasc Interv Neurol*. 2019;10(3):16-22. PMID:31308866.
4. Makowicz G, Poniatowska R, Lusawa M. Variants of cerebral arteries: anterior circulation. *Pol J Radiol*. 2013;78(3):42-7. <http://doi.org/10.12659/PJR.889403>. PMID:24115959.
5. Aso K, Kashimura H, Matsumoto Y, Saura H. Microsurgical clipping for anterior communicating artery aneurysm associated with the accessory anterior cerebral artery via the pterional approach. *Surg Neurol Int*. 2018;9(1):120. http://doi.org/10.4103/sni.sni_103_18. PMID:30009084.
6. Krzyżewski RM, Tomaszewski KA, Kochana M, Kopec M, Klimek-Piotrowska W, Walocha JA. Anatomical variations of the anterior communicating artery complex: gender relationship. *Surg Radiol Anat*. 2015;37(1):81-6. <http://doi.org/10.1007/s00276-014-1313-7>. PMID:24849465.
7. Narang S, Dil J, Raja A. Distal anterior cerebral artery aneurysms: a brief review. *Journal of Cerebrovascular Sciences*. 2022;10(1):35-40. http://doi.org/10.4103/jcvs.jcvs_18_22.
8. Al-Mufti F, Amuluru K, editors. *Cerebrovascular disorders*. Totowa: Humana Press; 2021. (Neuromethods; no. 170). <http://doi.org/10.1007/978-1-0716-1530-0>.
9. Secer M, Gokbel A. Surgical management and its outcomes in distal anterior cerebral artery aneurysms. *Int Med J*. 2021;10(4):1402. <http://doi.org/10.5455/medscience.2021.08.264>.

10. Bonasia S, Robert T. Retractorless combined pterional and interhemispheric approach to achieve proximal control in pericallosal artery aneurysm: how I do it. *Acta Neurochir*. 2021;163(10):2733-8. <http://doi.org/10.1007/s00701-021-04782-7>. PMID:33687560.

11. Xu K, Hou K, Xu B, Guo Y, Yu J. Single-Stage clipping of seven intracranial aneurysms in the anterior circulation via unilateral pterional approach: a case report and literature review. *J Neurol Surg A Cent Eur Neurosurg*. 2020;81(3):271-8. <http://doi.org/10.1055/s-0039-1698381>. PMID:31962354.

12. Kim M, Kim BJ, Son W, Park J. Postoperative clipping status after a pterional versus interhemispheric approach for high-positioned anterior communicating artery aneurysms. *J Korean Neurosurg Soc*. 2021;64(4):524-33. <http://doi.org/10.3340/jkns.2020.0215>. PMID:33853297.

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ERRATA: Aneurysmal Bone Cyst

Por um erro honesto dos autores, o artigo Aneurysmal Bone Cyst, publicado em J Bras Neurocir 36 (2):234-239, 2025 (DOI: <https://doi.org/10.22290/jbnc.2025.360203>), foi publicado com imagens incorretas. Para corrigir e esclarecer a pesquisa, as imagens e alguns trechos do documento foram ajustados e são apresentados nesta errata.

Aneurysmal Bone Cyst

Cisto Ósseo Aneurismático

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ABSTRACT

Introduction: *The Aneurysmal Bone Cyst (ABC) is a benign yet locally aggressive lesion primarily affecting the pediatric and young adult population. Initial assessment of an ABC requires X-rays, followed by Magnetic Resonance Imaging (MRI) or Computed Tomography (CT) for further examination.* **Case presentation:** *This case report illustrates an atypical presentation of the disease and sheds light on differential diagnoses and clinical nuances that indicated an unconventional course of disease. The case involved a male, 30 years old, previously healthy, whose chief complaint was progressive nasal obstruction for 6 months, and sporadic epistaxis. Notably, pain associated with these symptoms was absent. The patient received endoscopic nasal treatment to address the fractured sphenoid bone.* **Conclusion:** *The observed orbital displacement, septal deviation, and erosion of adjacent structures all underscored the need for a multidisciplinary approach to the case management.*

Keywords: Case report; Magnetic resonance imaging; Computed tomography; X-ray; Aneurysmal bone cyst

RESUMO

Introdução: O Cisto Ósseo Aneurismático (COA) é uma lesão benigna, porém localmente agressiva, que afeta principalmente a população pediátrica e adulta jovem. A avaliação inicial de um COA requer radiografias, seguidas de ressonâncias magnéticas (RM) ou tomografias computadorizadas (TC) para exame adicional. **Descrição do caso:** Este relato de caso ilustra uma apresentação atípica da doença e lança luz sobre diagnósticos diferenciais e nuances clínicas que indicaram um curso não convencional da doença. O caso envolveu um paciente masculino, 30 anos, previamente saudável, cuja principal queixa foi obstrução nasal progressiva por 6 meses, e epistaxe esporádica. Notavelmente, a dor associada a estes sintomas estava ausente. O paciente recebeu tratamento nasal endoscópico para tratar o osso esfenoidal fraturado. **Conclusão:** O deslocamento orbital, o desvio septal e a erosão das estruturas adjacentes observados ressaltaram a necessidade de uma abordagem multidisciplinar para o manejo do caso.

Palavras-Chave: Relato de caso; Ressonância magnética; Tomografia computadorizada; Radiografia; Cisto ósseo aneurismático

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INTRODUCTION

The Aneurysmal Bone Cyst (ABC) is a benign yet locally aggressive lesion primarily affecting the pediatric and young adult population. Initial assessment of an ABC requires X-rays, followed by MRIs or CTs for further examination. While imaging usually renders a variety of patterns, a common presentation involves a lytic and expansile lesion exhibiting fluid-fluid levels¹.

The pathogenesis of ABCs remains a subject of controversy, with proposed etiologies including vascular, traumatic, or genetic factors. In X-rays, these lesions manifest as dilated, radiolucent areas typically in the metaphyseal region of the bone and accompanied by discernible fluid-fluid levels as revealed by MRIs. Histologically, ABCs are also characterized by lake-islands in fibrous stromata. Noteworthy differential diagnoses include conditions such as telangiectatic osteosarcoma and giant cell tumors^{2,3}. Such findings in the nasal fossa require ongoing observation incorporating imaging, timely diagnosis, and comprehensive treatment strategies for enhancing patients' quality of life. The present case report elucidates an atypical presentation of ABC that offers insights into its diagnostic challenges and multifaceted management.

The cornerstone of treating ABCs is curettage and bone grafting, with or without adjuvant therapy. Alternatives include cryotherapy, sclerotherapy, and radionuclide ablation. The case presented here introduces endoscopic endonasal treatment as an innovative treatment for this disorder and demonstrates the evolving options in this field^{1,2}. These varied possibilities call for more research to refine our understanding and optimize patient outcomes.

CASE PRESENTATION

A male, 30 years old, previously healthy, presented as his chief complaint progressive nasal obstruction for 6 months, with sporadic epistaxis. Notably, there was an absence of pain associated with these symptoms.

A non-contrast-enhanced computed tomography (CT) of the paranasal sinuses, conducted in sagittal and coronal planes,

revealed an expansive lesion in the left nasal cavity. This lesion resulted in the destruction of the bony nasal septum, the left lateral nasal wall, erosion of the lamina papyracea, and involvement of the skull base along the anterior cranial fossa. The dimensions of the lesion were approximately 3.8 x 2.5 x 4.7 cm, with undetermined etiology and potential evolution. Additionally, there was mucosal thickening in the left maxillary and right frontal sinuses. Other paranasal cavities maintained normal aeration. The nasal septum exhibited a right convex curvature, with the formation of a small bony spur in the right middle meatus (Figures 1, 2, 3, 4).

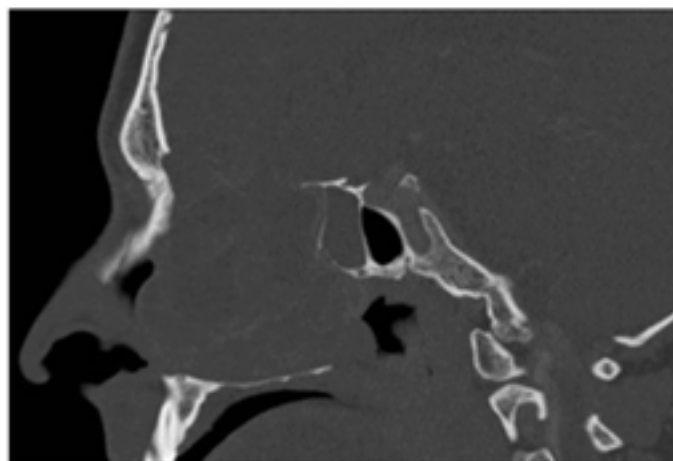


Figure 1. Preoperative sagittal bone window CT showing lesion in the nasal cavity.



Figure 2. Preoperative coronal bone window CT showing lesion in the nasal cavity.

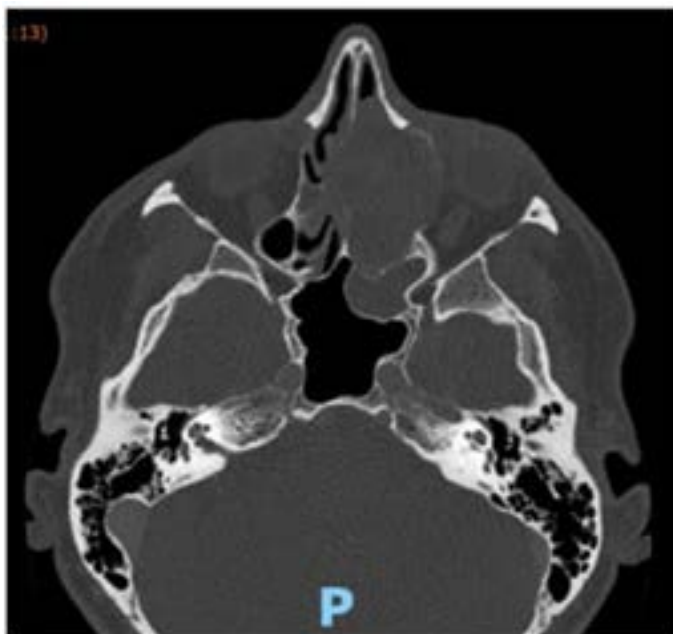


Figure 3. Preoperative axial bone window CT showing lesion in the nasal cavity.

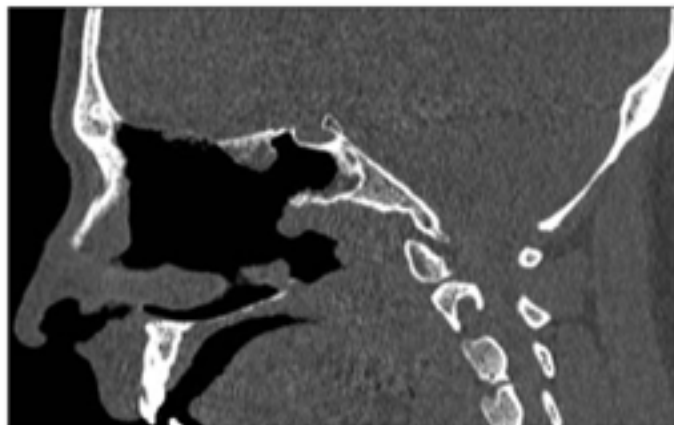


Figure 5. Postoperative sagittal bone window CT showing resection of the lesion.

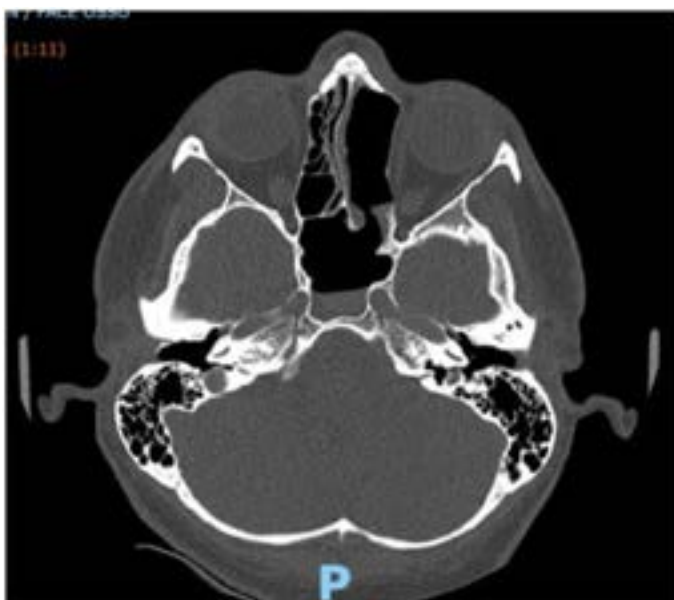


Figure 6. Postoperative axial bone window TC showing resection of the lesion.



Figure 4. Postoperative coronal bone window CT showing resection of the lesion.

Subsequent magnetic resonance imaging (MRI) of the paranasal sinuses, performed in axial, coronal, and sagittal planes with T1 and T2-weighted sequences (Figure 1), both with and without fat saturation, before and after Gadolinium

infusion on 26/7/2023, demonstrated a slight reduction in the dimensions of the expansive lesion. The lesion exhibited cystic characteristics involving the left ethmoid cells, with multiple loculations and fluid-fluid levels, suggestive of a mucocele. In the latest examination, the lesion measured approximately 4.8 x 3.8 x 2.2 cm, with an estimated volume of 21.01 cm³, compared to the previous measurement of 6.5 x 4.7 x 2.6 cm, with an estimated volume of 41.59 cm³ (Figures 4, 5, 6). Similar to the control examination, the lesion displayed peripheral enhancement in post-contrast sequences, extending into the ipsilateral nasal fossa. Hyperintense material in T1 suggested the

presence of hematic content or a high concentration of proteins. Furthermore, there was thinning of the lamina papyracea on the left, along with compression and displacement of the extrinsic orbital musculature. The lesion induced displacement of the nasal septum to the right, causing bulging and indistinctness of the ethmoidal roof. Despite the aeration of the left maxillary sinus, a small amount of mucus was retained. A fluid-air level appeared in the right frontal sinus, while the left frontal sinus showed agenesis. Other paranasal cavities maintained normal aeration, and the olfactory fossae had a depth of approximately 0.6 cm (Keros type II).

The macroscopic examination revealed two irregular portions of brownish and elastic tissue, measuring 3.7 x 2.8 x 0.9 cm. Histopathological analysis identified nasal mucosa and nasal bone with proliferative fibrous connective tissue forming membranes and cavities. The presence of fibrovascular stroma, hemosiderosis, chronic inflammatory infiltrate, osteoclast-like giant cells, and immature reactive bone was noted. Peripheral ossification was observed, along with nasal mucosa congestion and foci of hemorrhage. No mitoses or relevant cellular atypia were identified, and there was an absence of necrosis or signs of aggressiveness in native bone. The combined histopathological and radiological findings support the diagnosis of an aneurysmal bone cyst.

The endoscopic nasal approach was employed to treat the lesion. During the procedure, under general anesthesia, the patient was placed in a supine position with the head slightly elevated. Using a 0-degree endoscope, the surgeon carefully navigated through the nasal cavity to access the lesion. The lesion was fully visualized and debrided using microdebriders and suction. Careful dissection was performed to remove the cystic components while preserving surrounding structures. The lesion's attachment to the sphenoid bone was meticulously dissected to ensure complete removal. Hemostasis was achieved using bipolar cautery. Postoperatively, the nasal cavity was packed with absorbable material to reduce the risk of bleeding and promote healing. The patient was monitored for any immediate postoperative complications and was discharged with instructions for follow-up care, including saline irrigations and antibiotics to prevent infection. The use of an endoscopic approach minimized tissue trauma, reduced recovery time, and provided a clear visualization of the lesion, demonstrating the efficacy of this minimally invasive technique.

DISCUSSION

Radiographic manifestations of Aneurysmal Bone Cyst (ABC) exhibit heterogeneous characteristics, considering that these lesions may affect any osseous structure within the organism and any specific regions within a bone. Regarding long bones, principal differential diagnoses for ABC encompass Unicameral Bone Cyst (UBC), Giant Cell Tumor, and Telangiectatic Osteosarcoma. The latter, being a malignant bone neoplasm, presents the most significant diagnostic challenge, although its incidence is exceptionally rare in pediatric populations^{5,6}.

Definitive diagnosis of ABC necessitates systematic confirmation through biopsy, emphasizing identification of soft tissue components, given that telangiectatic sarcoma may simulate ABC. Intralesional sclerotherapy utilizing alcohol has demonstrated significant therapeutic efficacy. In cases involving spinal ABC and aggressive lesions with fracture risk, surgical intervention should be prioritized, potentially preceded by preoperative embolization. Risk of malignant transformation remains minimal, except in cases subjected to radiation therapy^{7,8}.

Concurrent utilization of non-contrast computed tomography and contrast-enhanced magnetic resonance imaging facilitated a comprehensive evaluation of the lesion. This multimodal approach provided detailed information regarding dimensions, characteristics, and potential involvement of adjacent structures. Association between macroscopic examination and histopathological analysis furnished detailed data concerning the nature of the lesion at a microscopic level, corroborating diagnosis of aneurysmal bone cyst and excluding aggressive characteristics.

The lesion's etiology remains indeterminate. Notwithstanding imaging and histopathological findings contributing to diagnosis of aneurysmal bone cyst, a more definitive understanding of an underlying cause or precipitating factors of the lesion has not been established. This case emphasizes the relevance of integrating clinical, radiological, and histopathological assessments for precise diagnosis. Volumetric reduction of the lesion over time suggests a dynamic nature, and the absence of aggressive characteristics in native bone constitutes a fundamental element for determining appropriate therapeutic strategy.

CONCLUSION

The clinical implications of this case extend beyond the diagnostic aspects, providing valuable insights into the potential progression and treatment response of aneurysmal bone cysts in the nasal cavity. The observed orbital displacement, septal deviation, and erosion of adjacent structures highlight the necessity for a multidisciplinary approach in managing such cases, integrating surgical expertise with a comprehensive understanding of the lesion's behavior. Careful observation of symptom onset and clinical presentation is crucial, as these factors, alongside laboratory tests, contribute to an accurate diagnosis and timely, appropriate treatment, thereby minimizing the risk of adverse outcomes.

REFERENCES

1. Restrepo R, Zahrah D, Pelaez L, Temple HT, Murakami JW. Update on aneurysmal bone cyst: pathophysiology, histology, imaging and treatment. *Pediatr Radiol*. 2022;52(9):1601-14. <http://doi.org/10.1007/s00247-022-05396-6>. PMID:35941207.
2. Mascard E, Gomez-Brouchet A, Lambot K. Bone cysts: unicameral and aneurysmal bone cyst. *Orthop Traumatol Surg Res*. 2015;101(1, Suppl):S119-27. <http://doi.org/10.1016/j.otsr.2014.06.031>. PMID:25579825.
3. Nasri E, Reith JD. Aneurysmal bone cyst: a review. *J Pathol Transl Med*. 2023;57(2):81-7. <http://doi.org/10.4132/jptm.2023.02.23>. PMID:36950810.
4. Rapp TB, Ward JP, Alaia MJ. Aneurysmal bone cyst. *J Am Acad Orthop Surg*. 2012;20(4):233-41. <http://doi.org/10.5435/JAAOS-20-04-233>. PMID:22474093.
5. Muratori F, Mondanelli N, Rizzo AR, et al. Aneurysmal bone cyst: a review of management. *Surg Technol Int*. 2019;35:325-35. PMID:31476792.
6. Pasquini E, Compadretti GC, Sciarretta V, Ippolito A. Transnasal endoscopic surgery for the treatment of fibrous dysplasia of maxillary sinus associated to aneurysmal bone cyst in a 5-year-old child. *Int J Pediatr Otorhinolaryngol*. 2002;62(1):59-62. [http://doi.org/10.1016/S0165-5876\(01\)00593-6](http://doi.org/10.1016/S0165-5876(01)00593-6). PMID:11738696.
7. Salmasi V, Blitz AM, Ishii M, Gallia GL. Expanded endonasal endoscopic approach for resection of a large skull base aneurysmal bone cyst in a pediatric patient with extensive cranial fibrous dysplasia. *Childs Nerv Syst*. 2011;27(4):649-56. PMID:21132434.
8. Yildirim AE, Ekici İ, Cagil E, Divanlioğlu D, Belen AD. Pure endoscopic endonasal removal of unusual anterior skull base aneurysmal bone cyst extending to the frontal lobe. *J Craniofac Surg*. 2015;26(3):e232-4. <http://doi.org/10.1097/SCS.0000000000001497>. PMID:25887207.

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