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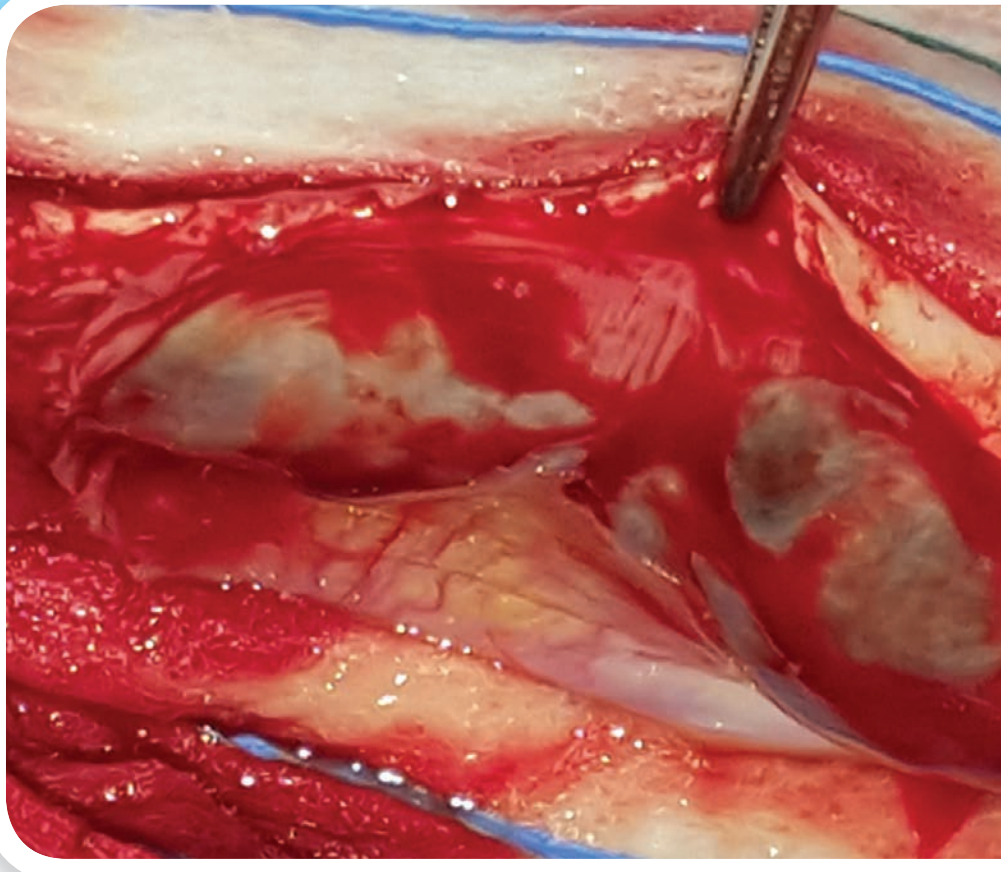

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Epidemiology and Estimated Cost of Surgery for Cubital Tunnel Syndrome Conducted by the Unified Health System in Brazil (2005–2015)

Epidemiologia e estimativa de custo das cirurgias para síndrome do túnel cubital realizadas pelo Sistema Único de Saúde no Brasil (2005–2015)

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Abstract

Introduction Cubital tunnel syndrome (CTS) is responsible for one of the types of ulnar nerve neuropathy and is the second cause of compressive neuropathy of the upper limb, only surpassed by carpal tunnel syndrome.

Objective To describe the epidemiological data of the ulnar nerve transposition surgical code in the treatment of CTS by the United Health System (SUS) from 2005 to 2015.

Methodology This is a descriptive epidemiological study, in which data were obtained through consultation of the DATASUS database.

Results/Discussion During this period, 774 procedures were performed and, despite the addition of 20.3 million people to the Brazilian population, the incidence was 0.33/1,000,000. National and international epidemiology point to a slightly higher prevalence of the procedure between men, in the fourth and fifth decades of life. Low permanence rate, as well as the absence of hospital deaths related to the procedure, infer that the procedure is safe, with low morbidity and mortality rates.

Conclusion The annual incidence of the cubital syndrome submitted to surgical treatment at SUS in the Brazilian population was 1/7,670,833 in 2005 and ½,174,468 in 2015. The cost of each surgical procedure during the same period ranged from R$ 318.88 to R$ 539.74. The mean hospitalization time for CTS surgery was 1.85 days.

Keywords
- epidemiology
- cubital tunnel syndrome
- ulnar nerve

Resumo

Introdução A síndrome do túnel cubital (STCB) é responsável por um dos tipos de neuropatia do nervo ulnar, sendo a segunda causa de neuropatia compressiva do membro superior, superada apenas pela síndrome do túnel do carpo.

Objetivo Descrever os dados epidemiológicos do código cirúrgico de transposição do nervo cubital no tratamento da STCB realizado pelo Sistema Único de Saúde (SUS), de 2005 a 2015.

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Metodologia Trata-se de um estudo epidemiológico descritivo cujos dados foram obtidos por meio de consulta à base de dados disponibilizada pelo DATASUS.

Resultado/Discussão Foram realizados 774 procedimentos ao longo deste período, e, apesar do acréscimo de 20,3 milhões de pessoas à população brasileira, constatou-se incidência de 0,33:1.000.000. A epidemiologia nacional e internacional aponta para uma discreta prevalência do procedimento em homens, entre a quarta e quinta décadas de vida. A baixa taxa de permanência e a ausência de óbitos hospitalares relacionados ao procedimento atestam que este é seguro, com baixa taxa de morbimortalidade.

Conclusão A incidência anual da STCB na população brasileira submetida ao tratamento cirúrgico, pelo SUS, em 2005, foi de 1:7.670.833, e em 2015, de 1:2.174.468. Foram gastos, para cada procedimento cirúrgico, de 2005 a 2015, valores que oscilaram entre R$ 318,88 e R$ 539,74. Observou-se média de 1,85 dias de permanência hospitalar para a realização da cirurgia da STCB.

Palavras-chave
- epidemiologia
- síndrome do túnel cubital
- nervo ulnar

Introduction

Cubital tunnel syndrome (CTS) is responsible for a type of ulnar nerve neuropathy and it is the second cause of compressive neuropathy of the upper limb, surpassed only by carpal tunnel syndrome. Since this nerve has a very long path in the upper limb, it can be compressed at several points, with the elbow region as the site of greatest impairment.[1,2]

Several etiologies are related to the development of CTS. These etiologies include metabolic alterations, congenital abnormalities, elbow trauma sequelae, tumors, osteoarthrosis and nerve subluxation in the medial epicondyle during elbow flexion.[3]

Epidemiological data show that its worldwide incidence is estimated at 25 cases per 100,000 people/year, with men being affected twice more than women, and with the highest impairment between the fourth and fifth decades of life.[1,2]

Italy presents the same worldwide incidence: 25 cases per 100,000 people/year.[4] In Germany, studies show an incidence of 24.7 cases per 100,000 people/year, involving 2 men for each woman.[5] In the Netherlands, the annual incidence is 21to 25 cases per 100,000 people.[6] In contrast, American epidemiological studies showed a higher incidence of CTS in women, around the 5th decade of life, affecting $\sim$ 376 per 100,000 people/year. Annually, 75,000 decompression surgeries are performed in the United States.[7,8]

There are few CTS-related studies in the Brazilian population regarding variables such as age, gender, ethnicity, occupation and clinical characteristics of the patients submitted to the surgical procedure.

Diagnosis is based on signs and symptoms, sensory and motor tests and electromyography of the upper limbs. The patients affected by this syndrome often present paresthesia in the ulnar nerve distribution territory, paresis and/or hypotrophy of the hand intrinsic musculature. The extent of ulnar nerve dysfunction is classified by McGowan into three grades: grade I, isolated sensory neuropathy; grade 2, sensory and motor neuropathy without muscular atrophy; grade 3, sensory and motor neuropathy with muscular atrophy.[2,9] Upper limb electromyography and clinical evaluation were considered the best parameters for the surgical decision making on CTS patients.[2]

Cubital tunnel syndrome treatment can be conservative or surgical, depending on the clinical signs and symptoms, according with the McGowan classification. Mild or moderate cases can be conservatively treated due to the potential of spontaneous regeneration. Surgical decompression is indicated in severe cases, or when there is no clinical improvement.[9]

Several surgical approaches are used in the treatment of ulnar neuropathy at the elbow, including simple decompression, endoscopic in situ decompression, ulnar nerve transposition (subcutaneous, intramuscular and submuscular), and medial epicondilectomy.[3,10,11] Although the best surgical technique is debated, the anterior transposition is the most used technique.[12]

Objective

To describe the epidemiological data regarding the number of annual procedures, hospital costs, hospitalization time and number of deaths of patients admitted by the Unified Health System (SUS), from 2005 to 2015, using the surgical code of ulnar nerve transposition.

Methodology

This is a descriptive epidemiological study with data obtained from the Information Technology Department of the Brazilian Public Health Care System (DATASUS, in the Portuguese acronym) database (http://www.datasus.gov.br) accessed in August and September 2016. The study population was composed of all patients submitted to ulnar nerve transposition (code 0403020107) from January 2005 to December 2015. The DATASUS data were tabulated using the SPSS 13.0 software (SPSS Inc., Chicago, IL, USA). Since

DATASUS is a public domain database, the project submission to the Research Ethics Committee was not required. Subsequently, a bibliographical survey was performed in scientific databases, searching for both Brazilian and international publications, in PubMed, BVS (*Biblioteca Virtual de Saúde*, Virtual Health Library), LILACS (*Literatura Latino-Americana e do Caribe em Ciências da Saúde*, Latin American and Caribbean Literature in Health Sciences), MEDLINE (National Library of Medicine) and SciELO (Scientific Electronic Library Online) with the following descriptors: epidemiology, cubital and syndrome. To narrow the search, only articles published between 2010 to 2016 were considered. The following inclusion criteria were used: indexed journals published in national and international journals, written in English and Portuguese.

The exclusion criteria considered papers whose titles and summaries did not fit the objectives of the research. From the defined strategy, the bibliographic search resulted in a final sample consisting of 20 publications, 8 indexed in the LILACS database, 5 in the MEDLINE database, and 7 in the SciELO database. The studies were carefully and completely read, and then selected for strictly meeting the inclusion criteria and deemed relevant to be part of the proposed study.

Results

►**Table 1** presents data on surgical procedures using the ulnar nerve transposition technique from January 2005 to December 2015. From the total of 774 procedures, 140 occurred in 2012, the year with the highest number of cases, representing 18.09% of the sample. There was an increase of 70 procedures between 2005 and 2015, representing an addition of 295.8%. In a comparative analysis of the number of procedures in this same period and the Brazilian population, it was observed that, even with the population increase, the annual incidence of patients submitted to surgery remained low, ranging from 1 case per 1,385,000 inhabitants to 1 case per 7,670,833 inhabitants (►**Table 2**).

Table 1 Total distribution of the number of ulnar nerve transposition procedures between 2005 and 2015 in the Unified Health System (SUS)

Year	Total AIH	%
2005	24	3.10
2006	42	5.43
2007	24	3.10
2008	64	8.27
2009	63	8.14
2010	65	8.40
2011	77	9.95
2012	140	18.09
2013	87	11.24
2014	94	12.14
2015	94	12.14
Total	774	100

Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

Table 2 Annual incidence of patients with cubital tunnel syndrome submitted to surgical treatment between 2005 and 2015 in the Unified Health System (SUS)

Year	Total AHS	Brazilian population (million)	Incidence
2005	24	184.1	1:7,670,833
2006	42	186.7	1:4,445,238
2007	24	183.9	1:7,662,500
2008	64	189.6	1:2,962,500
2009	63	190.7	1:3,026,984
2010	65	191.4	1:2,944,615
2011	77	192.3	1:1,497,402
2012	140	193.9	1:1,385,000
2013	87	201.0	1:2,310,344
2014	94	202.7	1:2,156,382
2015	94	204.4	1:2,174,468

Abbreviation: AHS, authorization for hospital stay.
Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

►**Table 3** shows the quantitative analysis by geographical region. Most surgeries occurred in the Southeast region, with 459 procedures, representing 59.3% of the total. In the Northern region, 18 surgical procedures were performed, the smallest amount among regions, corresponding to 2.32%. The total and average values of procedural costs, as well as hospital and professional costs, are shown in ►**Table 4**. The average hospital cost oscillated from R$ 315.23, the lowest value in 2005, to R$ 515.25, the lowest value in 2015, evidencing an increase of 63.44%. ►**Table 5** shows the data regarding the average number of hospitalization days, as well as the number of deaths. These data show a variation of 0.7 day in 2012 and 3.5 days in 2009, with an overall average of 1.85 days. No deaths occurred throughout the study period.

Table 3 Distribution of the number of ulnar nerve transposition procedures performed in the Unified Health System (SUS) between 2005 and 2015 per region

Region	Number	%
North	18	2.33
Northeast	65	8.40
Southeast	459	59.30
Central West	50	6.46
Total	774	100

Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

Table 4 Distribution of the actual costs, in Brazilian currency, of ulnar nerve transposition procedures performed in the Unified Health System (SUS) between 2005 and 2015 per region

Year	Total value	Average value	Hospital services cost	Professional services cost
2005	7,653.15	318.88	4,285.71	2,352.65
2006	13,568.02	323.05	7,616.77	4,094.25
2007	7,957.53	331.56	4,484.07	2,344.19
2008	28,017.92	437.78	17,389.13	10,628.79
2009	27,633.80	438.63	17,204.90	10,428.90
2010	28,770.11	442.62	17,645.43	11,124.68
2011	38,797.30	503.86	20,862.28	17,935.02
2012	72,725.95	519.47	37,963.39	34,762.56
2013	45,265.36	520.29	23,436.17	21,571.57
2014	52,059.84	553.83	27,390.03	23,381.67
2015	50,735.42	539.74	25,812.66	23,369.12

Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

Table 5 Mean distribution of hospitalization days and number of deaths related to ulnar nerve transposition procedures performed in the Unified Health System (SUS) between 2005 and 2015

Year	Average hospitalization days	Deaths
2005	2.1	–
2006	2.1	–
2007	1.8	–
2008	1.8	–
2009	3.5	–
2010	1.9	–
2011	1.4	–
2012	0.7	–
2013	1.2	–
2014	1.8	–
2015	2.1	–
Average	1.85	–

Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

►**Table 6** shows the distribution of the number of procedures per type/code performed for CTS treatment, as well as other codes that could be used for both CTS and other peripheral nerve compressive conditions. From 2005 to 2007, procedures were performed with the following codes: ulnar nerve transposition (procedure code: 0403020107) and neurolysis (procedure code: 40200043). Starting in 2008, three other codes have passed to be adopted: nonfunctioning neurolysis of peripheral nerves (code procedure: 0403020077), microneurolysis of peripheral nerve (procedure code: 0403020050) and surgical treatment of neuropathy compressive with or without microsurgery (procedure code: 0403020115).

Discussion

A total of 774 surgical procedures were performed using the ulnar nerve transposition technique (procedure code: 0403020107) from January 2005 to December 2015. This is considered the only specific code for the surgical treatment of the cubital tunnel syndrome available by SUS according to data obtained through the information system.

During the study period, there was a significant increase in the use of the ulnar nerve transposition technique, since 24 procedures were performed in 2005 and 94 procedures in 2015, showing a 295.8% increase.

The Brazilian population increased in 20.3 million individuals during the period studied. The annual incidence in 2005, when the estimated population was 184.1 million people, was 0.13:1,000,000, and, in 2015, when the population was 204.4 million, 0.46:1,000,000. The highest incidence was noted in 2012, with 0.72:1,000,000 inhabitants. Finally, the annual average incidence of cubital tunnel syndrome was 0.33:1,000,000.

A European study, performed at Centro Hospitalar do Tâmisa e Sousa, in Portugal, evaluated 36 patients (17 men and 19 women) diagnosed with CTS who underwent ulnar nerve transposition from 2006 to 2009. The mean age of the patients was 41.6 years old (23–72 years old). Of these, 78% of patients presented severe neuropathy and improved after surgery, with a satisfaction rate of 86%, and with 72% returning to daily activities without limitations.[3]

In Asia, a study conducted in Korea from 2010 to 2012, with 69 patients submitted to ulnar nerve transposition surgery, showed that their mean age was 36 years-old and there was a male prevalence (59%).[13]

In the United States, a study with 25 patients (14 men and 11 women) undergoing surgery between 2003 and 2009 observed that their mean age was 53 years-old. In this study, 20 patients were submitted to unilateral surgery and 5 to bilateral surgery.[14]

Table 6 Distribution of the number of procedures performed from 2005 to 2015 in the Unified Health System (SUS) specifically for cubital tunnel syndrome (CTS) treatment in addition to other procedures not specific for this condition

Year	Ulnar nerve transposition	Nonfunctional peripheral nerve neurolysis	Peripheral nerve microneurolysis	Surgical treatment*	Neurolysis	Total
2005	24	_	_	_	932	956
2006	42	_	_	_	1,181	1,223
2007	24	_	_	_	899	923
2008	64	3,123	4,376	5,151		12,714
2009	63	3,453	5,024	6,015		14,555
2010	65	3,157	4,874	5,988		14,084
2011	77	3,049	4,934	6,532		14,592
2012	140	3,291	5,059	7,139		15,629
2013	87	3,021	4,821	7,156		15,085
2014	94	3,134	4,918	7,064		15,210
2015	94	3,917	6,413	8,508		18,932
Total	774	26,145	40,419	53,553	3,012	123,903

*surgical treatment of compressive neuropathy with or without microsurgery.
Source: http://www2.datasus.gov.br/DATASUS/index.php?area=02.

In a Brazilian study performed between 2001 and 2006 at the Institute of the Hand of Universidade Federal de São Paulo (Unifesp) with 21 CTS patients treated with ulnar nerve transposition surgery, 12 individuals (57.1%) were male. The mean age of the patients was 51.6 years old.[15] Still in Brazil, at Hospital Sarah Brasília, 58 cases were studied between 2001 and 2007 and 6 cases were bilaterally operated. In this study, a female predominance and the preponderance of the 40 to 50 years old age group were observed.[16]

Based on the above-mentioned studies, it can be inferred that the national and international epidemiology points to a discrete prevalence of the ulnar nerve transposition procedure in men between the fourth and fifth decades of life. However, data on annual prevalence and incidence, as well as hospital costs, are still scarce.

The quantitative analysis by region showed the following: the North region had the lowest number of surgeries (18), while the Southeast region had the largest number (459), accounting to 59.30% of the total. These data suggest that some factors could justify this difference, such as population density, work activity type, access to health care and the ability of the medical professional to recognize the condition.

According to DATASUS, R$ 7,653.15 were spent on CTS surgeries in 2005, with an average cost of R$ 318.88 per procedure. In 2015, the total expense was R$ 50,735.42, or R$ 539.74 per procedure, showing an increase of 69.25% in the average hospital cost of this surgery. It is noteworthy that, according to SUS, R$ 432.60 were paid per procedure between 2008 and 2011, with an increase to R$ 515.25 starting on 2012, corresponding to a 19.1% increase.

Regarding the mean number of hospitalization days, also defined as the minimum hospitalization rate, we observed an oscillation between 0.7 days in 2012 to 3.5 days in 2009, with an overall mean of 1.85 days of hospitalization associated to the surgical procedure. It is noteworthy that the death rate during the study period was zero.

It is important to consider that some segments were excluded from the analysis proposed by this study: patients coming from the private health network; CTS patients submitted to clinical treatment, with no need for surgical procedure; and CTS patients who underwent surgical treatment using other codes available at SUS (neurolysis, nonfunctioning peripheral nerve neurolysis, peripheral nerve microneurolysis and compressive neuropathy treatment with or without microsurgery). These last four codes totaled 120,117 procedures during the studied period. From the total above, it is possible to infer that there is a procedural fraction not quantified for CTS. As such, it is possible to affirm that the present study data could be underestimated.

Conclusion

The annual incidence of cubital tunnel syndrome in the Brazilian population submitted to surgical treatment in the SUS between 2005 and 2015 was 0,33:1,000,000. Each surgical procedure cost between R$ 318.88 and R$ 539.74 during this period. The mean hospitalization time for CTS surgery was 1.85 days. The death rate was null during the study period. New epidemiological studies should involve the population coming from the private health sector as well as clinically treated CTS patients.

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Brachial Plexus Injuries with Ulnar Musculocutaneous Transfer: 78 Cases Study Focused on Possible Postoperative Complications

Transferência ulnarmusculocutânea nas lesões de plexo braquial: estudo de 78 casos quanto a possíveis complicações pós-operatórias

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Abstract

Objective To analyze 78 cases of brachial plexus injury submitted to the Oberlin technique between 2003 and 2012. The potential complications of this technique were analyzed, especially motor damage or hypoesthesia of the hand.

Method Medical records from patients with brachial plexus injuries at the levels of the C5-C6 and C5-C6-C7 vertebrae were retrospectively analyzed. Cases submitted to the Oberlin procedure with or without concomitant brachial plexus procedures between 2003 and 2012 were evaluated. The minimum follow-up period was of 1 year. In addition to the clinical examination, electromyography and magnetic resonance imaging (MRI) of the brachial plexus were used to diagnose and locate the nerve damage.

Results A total of 78 surgical patients met the inclusion criteria. Postoperative neurological changes, mostly transient, were observed in 18 patients. Hypoesthesia in the ulnar side of the hand was observed in seven cases; neuropathic pain in five cases; allodynia in four cases, and hand motor loss in two cases.

Conclusion Based on the results of the present case series, we conclude that there are few sequelae in the donor nerve territory compared with the benefit of the Oberlin technique on the recovery of elbow flexion after brachial plexus injuries.

Keywords
- nerve transfer
- ulnar nerve
- musculocutaneous nerve

Resumo

Objetivo Analisar 78 casos de lesão de plexo braquial operados, submetidos à técnica de Oberlin entre 2003 e 2012. Possíveis complicações desta técnica foram analisadas, sobretudo possíveis prejuízos motores ou hipoestesia na mão.

Método Foi realizada uma análise retrospectiva de prontuários de pacientes com lesões do plexo braquial com lesão nos níveis das vértebras C5-C6 e C5-C6-C7. Foram analisados casos que haviam sido submetidos ao procedimento de Oberlin associado ou não a outros procedimentos concomitantes do plexo braquial entre 2003 e 2012. O

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seguimento pós-operatório mínimo foi de 1 ano. Além do exame clínico, foram usadas para o diagnóstico e para a localização da lesão nervosa a eletroneuromiografia e a ressonância nuclear magnética do plexo braquial.

Resultados Foram obtidos 78 casos operados que preenchiam os critérios de inclusão. Alterações neurológicas pós-operatórias foram observadas em 18 pacientes, sendo, em sua grande maioria, transitórias. Hipoestesia na face ulnar da mão foi observada em sete casos; dores neuropáticas, em cinco; alodinia, em quatro; e perda motora na mão, em dois casos.

Conclusão Considerando os resultados da nossa casuística, concluímos que as sequelas no território do nervo doador são poucas diante do benefício que a técnica de Oberlin pode trazer à recuperação da flexão do cotovelo nas lesões do plexo braquial.

Palavras-Chave
- transferência nervosa
- nervo ulnar
- nervo musculocutâneo

Introduction

The frequency of brachial plexus injuries has increased progressively, mostly due to motorcycle accidents.[1] The roots of the C5 and C6 vertebrae are often affected, resulting in neurological deficits in the shoulder and elbow joints, but sparing hand function.[2] Direct repairs with grafts are feasible only in postganglionic nerve lesions, while nerve transfers (neurotizations)[2,3] are indicated for preganglionic lesions.

Until the early 1990s, the nerve transfer procedures for the treatment of upper brachial plexus roots lesions involved accessory nerves or intercostal nerves.[4,5] Oberlin et al published an intraplexual nerve transfer technique using one or two ulnar nerve fascicles with terminoterminal neurorrhaphy to the biceps motor branches in an attempt to restore elbow flexion.[3] Mackinnon et al described a variant of the original Oberlin technique in which, in addition to the ulnar-musculocutaneous transfer, a bundle of the median nerve was transferred to the motor branch for the brachial muscle.[6,7]

The Oberlin technique with the ulnar nerve as a donor has been shown to be simple to execute, and it is associated with few risks of severe sequelae due to the section of donor fascicles.[8,9] However, there is a concern not to increase morbidity by using a functional donor nerve for the hand. Therefore, the present study aims to describe our experience with the original Oberlin technique, emphasizing its postoperative morbidity and the safety of the surgical approach.

Method

Medical records from patients submitted to the Oberlin procedure as surgical treatment of brachial plexus lesions involving the C5-C6 and C5-C6-C7 vertebrae were reviewed. The present study analyzed patients treated between 2003 and 2012, and with at least 1 year of postoperative follow-up. The variables analyzed included hand muscle strength and the presence of hypoesthesia, of sensitivity changes, and of neuropathic pain. These evaluations were performed by an independent examiner and correlated with factors such as the age of the patient age at the time of surgery, time elapsed between the trauma and the procedure, and the number of ulnar nerve fascicles used in the nerve transfer. The postoperative follow-up by the rehabilitation team occurred 30 days, 6 months, and 1 year after the discharge and, subsequently, annually. The functional assessment was performed according to the International Classification of Motor Function (Veterans Administration), detailed in ►**Table 1**. Other procedures performed in these patients for the restoration of shoulder function, such as accessory nerve transfer to the suprascapular muscle, triceps motor branch transfer to the axillary nerve, and graft reconstructions, were not considered in the present study.

The surgical technique used was described by Oberlin (►**Fig. 1**).[3] The patient was placed in the supine position with the affected upper limb in abduction. An incision was then made in the midline of the medial portion of the arm, between its middle and upper third. The space between the brachialis and the coracobrachialis biceps muscles was explored, identifying the motor branch to the biceps, which arises from the musculocutaneous nerve.

Next, the ulnar nerve was identified medially to the brachial artery, and the anterolateral aspect of its epineurium was incised (►**Fig. 2**). The motor fascicles for the flexor carpi ulnaris muscle were identified by a nerve microstimulator.

One or two motor fascicles were then sectioned, and a terminoterminal neurorrhaphy was performed between these ulnar nerve fascicles and the musculocutaneous branch to the biceps brachii using monofilament nylon 9.0, which can be reinforced with fibrin glue. After performing the suture, a test

Table 1 Motor function international classification (Veterans Administration)

M0	No motor activity
M1	Visible or palpable muscle contraction, but with no active movement
M2	Motor function present when gravity is eliminated
M3	Active movement overcomes gravity, but not resistance
M4	Active movement overcomes resistance
M5	Normal muscle strength

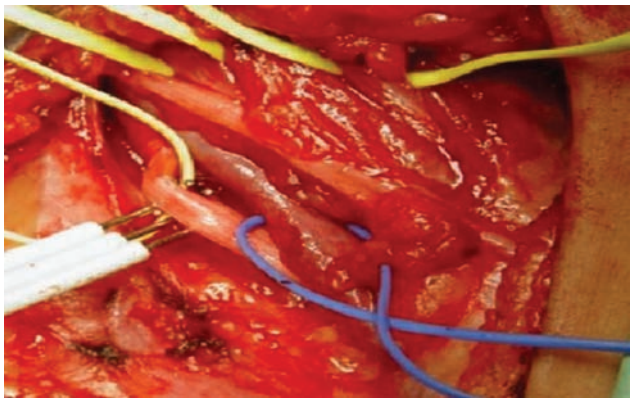

Fig. 1 Approach to the nerves of the arm. The ulnar nerve is observed medially, with the nerve stimulator, and the musculocutaneous nerve laterally, evidenced by the yellow strip.

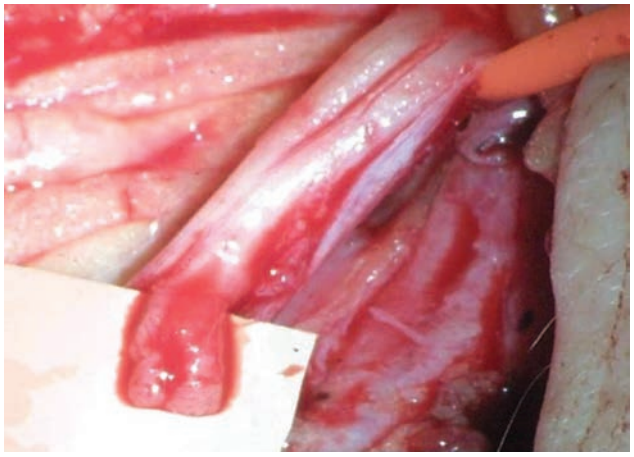

Fig. 2 The ulnar nerve epineurium was opened, and two fascicles were prepared and sectioned after the stimulation.

performed by moving the upper limb assured the absence of tension. Finally, the upper limb was immobilized in a sling for ~ 30 days.

Results

A total of 78 patients were analyzed, of which 74 were male. Children with obstetric brachial palsy and < 12 years old with traumatic injuries were excluded from the study, mainly due to their difficulty to report sensitivity changes and pain characterization. The age of the patients at the time of the trauma ranged from 21 to 52 years old, and the average time elapsed until surgery was 7 months (ranging from 3 to 15 months). There were no intraoperative complications, such as dehiscence or infection of the surgical area. All of the patients were properly immobilized during the postoperative period. Postoperative complications related to the ulnar nerve included hypoesthesia in the ulnar face of the hand, neuropathic pain, allodynia, and loss of muscle strength in the hand. These changes, mostly transient, were observed in 18 patients (23% of the sample), all male (►**Table 2**).

Among the seven cases of hypoesthesia of the ulnar side of the hand, three remained without remission, that is, sustaining some degree of the condition. Among the 4 cases that showed improvement, 2 recovered in < 6 months postoperatively, one recovered after 7 months, and the other case took > 1 year to achieve complete remission (►**Table 3**). From the three cases without remission, two used one ulnar nerve fascicle for nerve transfer, and the third case used two fascicles. In 2 cases, the patient was operated 8 months post-trauma, and, in 1 case, 5 months had elapsed since the traumatic event. In the case with hand sensitivity remission after > 1 year post-surgery, 2 fascicles had been used, and the time of evolution of the lesion at the time of the surgery was 6 months. There was no statistical correlation between the number of fascicles used and the occurrence of hypoesthesia.

Regarding neuropathic pain, only 1 case remained without remission, another 3 improved in < 6 months, and 1 case presented complete recovery after > 1 year (►**Table 4**). Patients with pain were all male, aged between 22 and 41 years old at the time of the surgery, and, in all of the cases, an ulnar nerve bundle was used for nerve transfer. In the case with no remission, 1 fascicle had been used, and the time elapsed between the lesion and the surgery was 6 months.

Complaints of allodynia on the ulnar side of the hand persisted in only 1 patient after the surgery, and 3 cases presented complete remission in < 6 months postoperatively (►**Table 5**). The age of the patients at the time of the surgery ranged from 21 to 36 years old; one of them was female; and all of the cases used an ulnar nerve bundle for the transfer. In the case that presented with sequelae, with no remission, the time elapsed between the trauma and the surgery was 8 months. Finally, there were 2 cases of wrist flexion and of digital deep flexion motor loss, but both were partial and transient, with

Table 2 Postoperative complications related to the ulnar nerve

Complication	Number of cases	Percentage
Ulnar face hypoesthesia	7	8.9%
Neuropathic pain	5	6.4%
Allodynia	4	5.1%
Motor loss in the hand	2	2.6%

Table 3 Hypoesthesia on the ulnar face of the hand

Postoperative recovery time	Number of cases
Without remission	3
< 0 6 months	2
7 months–1 year	1
> 1 year	1

Table 4 Neuropathic pain

Postoperative recovery time	Number of cases
Without remission	1
< 6 months	3
> 1 year	1

Table 5 Alodinia

Postoperative recovery time	Number of cases
Without remission	1
< 6 months	3

complete remission in ~ 30 days post-surgery. The first patient, male, was 36 years old at the time of the trauma, and only 1 ulnar nerve bundle was used in the nerve transfer surgery, 60 days after the accident. The second patient, also male, was 27 years old, and only 1 ulnar nerve fascicle was used in the surgical transfer, 7 months post-trauma. Muscle strength was recovered 40 days post-surgery.

Therefore, we have noticed that most of the complications in our series were transient, requiring < 6 months for complete recovery.

Discussion

Nerve transfers modified the approach to peripheral nerve lesions, especially to brachial plexus lesions.[2] Transfers can use extraplexual or intraplexual donor nerves; in partial lesions, the plexus itself is used as a donor to the injured nerves.[2,10,11] Nerve transfers to treat brachial plexus high lesions (at the levels of the C5-C6 and of the C5-C6-C7 vertebrae) were compared by Garg in a systematic review.[12,13] The 31 analyzed studies demonstrated the superiority of nerve transfer compared with the direct approach to the brachial plexus with grafts.[13]

The literature reports excellent outcomes for the nerve transfer surgery for the recovery of elbow flexion.[12,14–16] Rezende et al, analyzing 19 patients with lesions at the levels of the C5-C6 and of the C5-C6-C7 vertebrae submitted to the Oberlin procedure, noted that 8 patients showed improvement to grade 4 muscle strength, but with changes in the hand after surgery.[16] These authors pointed out that the time elapsed between the lesion and the surgery was the main prognostic factor for functional recovery.[16] A study conducted by a group from the Universidade de São Paulo and from the Universidad de Buenos Aires compared the nerve transfer with grafting and found that the former is associated with a superior outcome regarding muscular strength.[11] Several authors consider that the great advantages of the Oberlin technique include promoting an effective motor response, not using the sural nerve as a graft, not sacrificing completely the donor nerve, and the proximity between the suture and the muscle requiring reinervation.[2,11,17] Ulnar-musculocutaneous transfer surgery uses only one or two ulnar nerve fascicles, and no significant functional deficit has been observed.[3,11] Few studies analyzed in detail the post-surgical deficits.[5,15] The good outcomes associated with the Oberlin technique are due to its simplicity and to the proximity between the suture and the muscle requiring reinnervation; in addition, these outcomes were reproduced by several different authors.[3,4,9,15]

The surgical complications of this technique are few and, therefore, were barely studied in the reported case series. Concerns regarding the preservation of hand muscle strength, sensory deficit, and the risk of neuropathic pain are the main neurological complications researched in this surgery.[3,18]

Intraoperative nerve stimulation avoids the random selection of ulnar nerve fascicles and is an essential aid to preserve fascicles with functional importance to the hand.[7,8,18] The use of stimulation prevents motor deficit, but with no protective action against significant sensorial deficit or even against the development of neuropathic pain, which theoretically can be extremely disabling for the patient. A study with 36 patients, performed by Sungpet, showed no change in hand or wrist strength or sensitivity.[19] This author used only 1 ulnar nerve fascicle and demonstrated a strength recovery to grade 3 in 34 patients with reinnervation starting at 3.3 months postoperatively.[18]

The use of the ulnar nerve as a donor has also been described in obstetric brachial palsies, in which there is a concern with the adequate development of the hand of the child.[4,5] A study by Siqueira et al analyzed the growth of the hand and applied the Al-Quattan scale to evaluate the manual function of children submitted to the procedure.[5] This study, in addition to demonstrating the efficiency of the surgical procedure, evidenced that the operated hand presented the same bone development compared with the unoperated one. Moreover, the Al-Quattan scale results were similar to the ones obtained before the surgery, proving that there was no motor deficit in the hands of children.[5] This study is one of the few in the literature to prospectively analyze the risk of deficit development after sectioning the ulnar nerve in order to use it as a donor.[5]

In the present case series, as has already been stated, only two cases presented with motor loss, which was transient. A few cases had some type of permanent sequela (three cases of hypoesthesia on the ulnar side of the hand, one case of neuropathic pain, and one case of allodynia), all of which were sensorial. These data lead us to conclude that the Oberlin technique has a low morbidity and, therefore, is a safe technique for the patient, confirming the data found in the literature review. The present study is limited due to the fact that it is a retrospective analysis of surgical cases. Although hand motor function and sensitivity were analyzed by an independent examiner, since the present study is a retrospective analysis, especially in terms of sensitivity, it can provide a lower frequency if compared with a prospective study with an active search directed to sensitivity.

We conclude that the Oberlin technique is already classically recognized as effective for improving the elbow flexion function in high brachial plexus lesions, with a low risk of morbidity.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Full-Videoendoscopic Interlaminar Surgery for the Treatment of Lumbar Disc Herniations—A Series of 50 Cases

Cirurgia totalmente videoendoscópica interlaminar para tratamento de hérnia de disco lombar—série de 50 casos

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Abstract

The present study aims to describe the results of full-videoendoscopic surgery through the interlaminar route for central lumbar disc herniation in a series of 50 cases in Brazil.
This is a retrospective single-center study. With the aim of describing safety, the present study reports the complication and revision rates. The clinical results were collected with the visual analogue scale (VAS) and with the Oswestry Disability Index (ODI) (a questionnaire to evaluate functional disability) at the preoperative visit and at 6 months postsurgery.
The average surgical time was 20 minutes (range: 9–40 minutes), and 100% of the procedures were performed in an outpatient setting. The mean VAS scores improved from 9.4 to 1.1 ($p < 0.001$), and the mean ODI scores decreased from 69 to 9 points in the last follow-up ($p < 0.001$). There was 1 case (2%) with hernia recurrence, 1 case with intraoperative root injury (2%), and 2 cases (4%) that required lumbar fusion due to a preexisting instability. No infections were observed.
The full-videoendoscopic surgery is a modern option for treating lumbar disc herniation. In the present study, we have observed that the use of this technique for the removal of fragments affecting the vertebral canal presented satisfactory clinical results, low complication rates, and that it has demonstrated its feasibility in an outpatient setting without prolonged hospitalization.

Keywords
- endoscopy
- intervertebral disc displacement
- minimally invasive surgical procedures
- lumbar spine

Resumo

O objetivo do presente estudo é descrever os resultados do uso da cirurgia totalmente videoendoscópica para a retirada de hérnias centrais, utilizando a rota interlaminar em uma série de 50 casos no Brasil.
Estudo não comparativo de série de casos com análise retrospectiva de dados coletados prospectivamente em um único centro. Com o intuito de descrever a segurança, foram relatadas as complicações e também as reoperações e recidivas

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observadas durante o seguimento. Para a realização da análise de resultados clínicos, foram utilizados como instrumentos a escala visual analógica (EVA) e o índice Oswestry de incapacidade (ODI, na sigla em inglês) (questionário para a quantificação de incapacidade funcional) antes e 6 meses após a cirurgia.
O tempo médio de cirurgia foi de 20 minutos (entre 9 e 40 minutos), com 100% dos pacientes tendo alta hospitalar no mesmo dia do procedimento. A média da EVA melhorou de 9,4 para 1,1 ($p < 0,001$), e a média do ODI caiu de 69 para 9 pontos no último acompanhamento ($p < 0,001$). Houve recidiva de hérnia em 1 caso (2%), 1 lesão intraoperatória (2%) de raiz, e necessidade de artrodese lombar em 2 casos (4%). Não ocorreu nenhum caso de infecção.
A cirurgia totalmente videoendoscópica é uma opção moderna para a retirada de hérnia de disco lombar. No presente trabalho, vimos que a utilização desta técnica para a retirada de fragmentos acometendo o canal vertebral gerou ótimos resultados de resolução clínica, com baixas complicações, e demonstrou ser factível em modelo ambulatorial sem internação hospitalar prolongada.

Palavras-chave
- endoscopia
- deslocamento do disco intervertebral
- procedimentos cirúrgicos minimamente invasivos
- coluna vertebral

Introduction

Open discectomy procedures evolved into percutaneous microdiscectomies since the adoption of microscopy, described by Yasargil in 1967,[1] and into other variations described by Hijikata et al in 1975[2] and by Caspar in 1977.[3] Since then, less invasive variations in spine surgical techniques have been implemented to decrease perioperative morbidity and postoperative infections, as well as to prevent damage to adjacent tissues. In addition, these strategies have direct and measurable effects on perioperative outcomes, on mobilization, on hospitalization, on return to activities and, finally, on societal costs.[4,5]

Today, less invasive, endoscopic procedures are performed in central[6,7] and in peripheral neurosurgery.[8] Moreover, these techniques are becoming well-established in lumbar disc herniation surgery. In cases presenting with disc fragments affecting the foramen or the lateral recess, the posterolateral endoscopic procedure[9,10] is performed by the transforaminal route through the Kambin triangle, or by the extraforaminal route. Cases with intracanal fragments are more challenging, and these routes are not used in them. Thus, the interlaminar route employed in full-videoendoscopic surgery has been recently described and has been used for disc herniations occupying the area of the vertebral canal.[11–13]

The present study aims to describe the results of the full-videoendoscopic surgery in the removal of central hernias using the interlaminar route in a Brazilian case series of 50 cases.

Materials and Methods

This is a non-comparative study of a case series with retrospective analysis of data prospectively collected in a single center from March 2014 to March 2016. The present study was analyzed and approved by the ethics committee of the institution in which it was conducted.

Inclusion/Exclusion Criteria

The inclusion criteria for the present study were: cases with central or centrolateral lumbar disc herniation diagnosed by magnetic resonance imaging (MRI); lack of clinical success after between 3 and 6 weeks of conservative treatment or cases evolving with acute neurological deficit; cases with symptomatology of unilateral radiculopathy; cases submitted to interlaminar, full-videoendoscopic surgery; minimum surgical follow-up of 6 months. The exclusion criteria were: cases with herniated lumbar disc in the foraminal or in the extraforaminal regions; cases treated endoscopically by the transforaminal or by the extreme lateral route; isolated lumbar pain; vertebral canal stenosis; presence of a facet cyst; discitis; cauda equina syndrome.

Surgical Technique

All of the surgical procedures were performed by the same neurosurgeon. The surgical procedure is illustrated in ►**Fig. 1**, and the assembly of the instrumentation table is shown in ►**Fig. 2**. The procedures were performed under general anesthesia. The surgical target in the present study was the posterior portion of the intervertebral disc through the interlaminar window (►**Fig. 1A-D**). The interlaminar videoendoscopic technique[11–13] is performed with a paramedian skin incision close to the median line, which is previously marked by fluoroscopy; then, a 6.9 mm diameter dilator is inserted toward the supraligamental interlaminar window (►**Fig. 1E-F**). After the insertion of the dilator, a 7.9 mm diameter working channel is placed and its bevel is directed to the ligamentum flavum; this procedure is aided by fluoroscopy (►**Fig. 1G**). The positioning of the endoscope through the working channel (►**Fig. 1H**) allows the visualization of the adjacent structures; the ligamentum flavum is opened with scissors, using the beveled work channel to retract the adjacent neural structures under direct endoscopic vision. The endoscopic view with 6.9 × 5.6 mm optics, 165 mm length and 25° angulation (►**Fig. 1I**) allows the

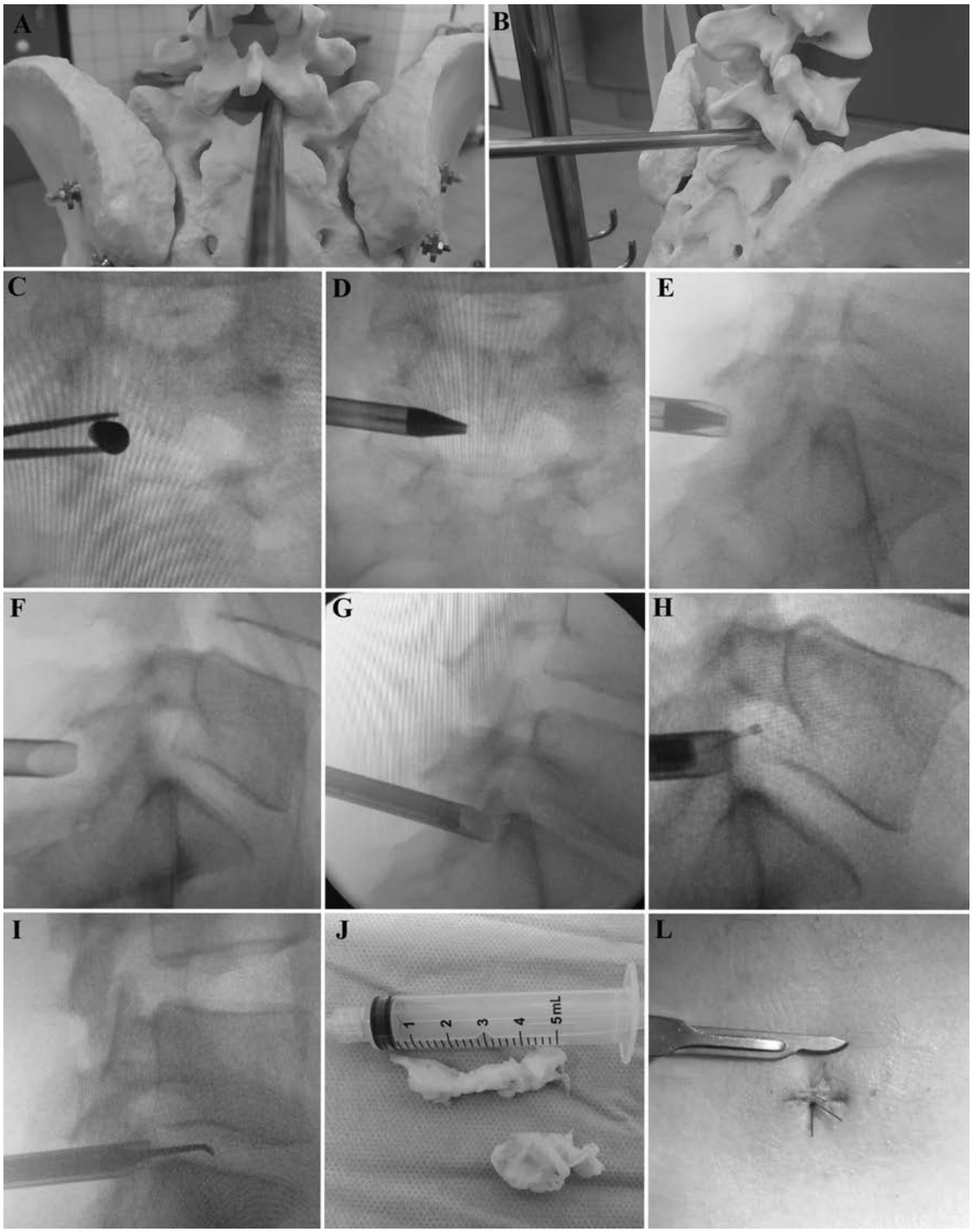

Fig. 1 Full-videoendoscopic surgical procedure through the interlaminar route. An anatomical model exemplifies (A) the anteroposterior view and (B) the lateral view. The surgical procedure is controlled by fluoroscopy for (C-E) localization, (F) interlaminar route access, (G and H) channel access, and (I and J) the removal of disc fragments. (L) The surgical incision is closed with a skin suture.

detailed observation of the structural intimate relationship (between the herniated fragment, the bulgings, the root of the nerve, and of the joint and the lamina of the facet). The use of appropriate materials, such as dissector, disc clamp, punch, and radiofrequency bipolar cautery, allows the performance of the procedure with greater safety. The intervertebral disc herniated fragment(s) is (are) removed (►**Fig. 1J**), and the procedure is finalized with rigorous hemostasis, local steroid placement, and skin suture using nylon 3.0 (►**Fig. 1L**). In order to avoid infections, the following protocol is used: cefuroxime 2 g intravenously during the anesthetic induction and cephalexin at a therapeutic dose for 7 days postoperatively.

The surgery described above is planned with a day hospital protocol, without the need for an overnight stay. In this protocol, the patient is admitted 3 hours before the

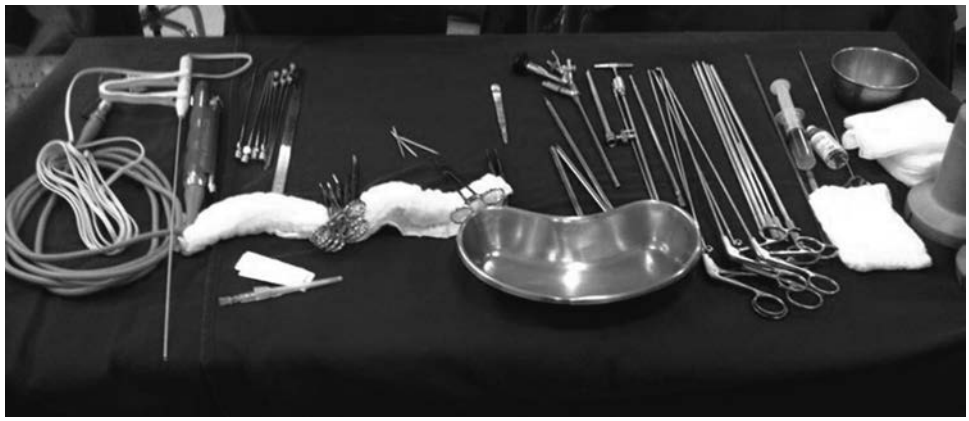

Fig. 2 Exemplification of the surgical materials used and of the configuration of the instrument table.

procedure and is discharged after 3 hours of postanesthetic observation, on the same day of the procedure.

Analyzed Outcomes

Data were collected before the surgery, intraoperatively, and 6 months after the procedure. Intraoperative and postoperative complications, as well as revision surgeries, were recorded.

The following instruments were used to analyze the clinical improvement after the surgery: the visual analogue scale[14] (VAS) for lumbar and irradiated pain (for pain assessment) and the Oswestry Disability Index[15] (ODI) (for functional disability assessment). The patients completed these questionnaires before the procedure and 6 months postsurgery.

Statistical Analysis

The data were submitted to a qualitative and quantitative descriptive analysis and to a comparative statistical analysis. The statistical tests were performed with SPSS Statistics for Windows, Version 10.0 (SPSS Inc., Chicago, IL, USA) using α values of 0.05. The two-tailed paired Student *t*-test was performed.

Results

Study Group

From a total of 123 patients submitted to videoendoscopic surgery at the spinal surgery center, 73 were cases treated through the posterolateral or the extraforaminal routes. The present study analyzed 50 cases that met the inclusion and exclusion criteria. ►**Table 1** shows the demographic data of the group. From the 50 selected patients, 20 (40%) were female, and 30 (60%) were male, with ages ranging from 19 to 69 years old, with a mean age of 32.4 years old. Regarding lumbar disc involvement, 1 (2%) was at the L3-L4 lumbar level; 23 cases (46%), L4-L5; and 26 (52%), L5-S1. The clinical picture lasted from 2 days to 13 months.

Surgical Results

Surgery data are shown in ►**Table 2**. The mean time for the surgical procedure was 20 minutes (9–40 minutes). There was no significant blood loss in any case, and, thus, there was no need for blood transfusion. There was no inadvertent durotomy during the analyzed procedures. With a single suture at the surgical incisions, no overnight hospitalization was required, and the median hospital stay was 3 hours.

Table 1 Demographic data

Total cases	50
Female	20 (40%)
Male	30 (60%)
Age (years old)	32.4 (19–69)
Time with preoperative symptoms (months)	8 (2–13)
L3-L4 lumbar level	1 (2%)
L4-L5 lumbar level	23 (46%)
L5-S1 lumbar level	26 (52%)

Clinical Improvement

Pain and physical disability levels, determined respectively by the VAS and ODI scales, were compared before and after the surgery (6 months after the procedure). In the pain scale, an improvement of 88% was observed at 6 months, with a statistically significant reduction from the original average score of 9.4 to 1.1 ($p < 0.001$) (►**Fig. 3**). Physical disability

Table 2 Surgical and postoperative data

Surgical time (minutes)	20 (9–40)
Durotomy	0 (0%)
Intraoperative lesion in L5	1 (2%)
Infection	0 (0%)
Hospitalization (hours)	3 (2.5–4)
Herniation recurrence	1 (2%)
Review (arthrodesis)	2 (4%)

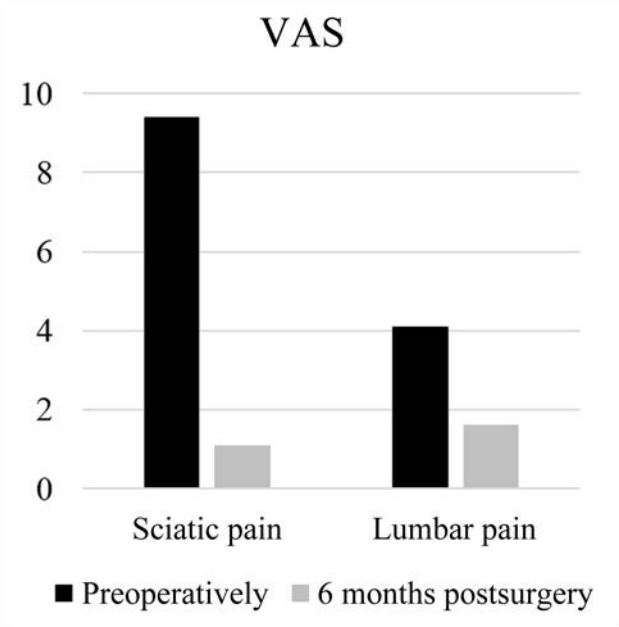

Fig. 3 Mean pain assessed by the visual analogue scale (VAS) in the studied group before the procedure and 6 months postsurgery ($p < 0.001$).

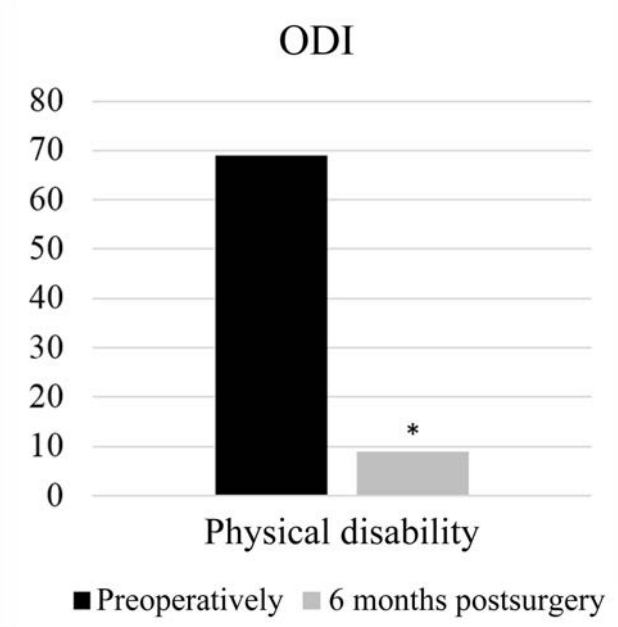

Fig. 4 Mean of the physical inactivity score measured by the Oswestry disability index (ODI) in the studied group before the procedure and 6 months postsurgery * $p < 0.001$.

showed an 87% decrease, from 69 to 9 points in the final follow-up ($p < 0.001$) (►**Fig. 4**). An exemplificative case is shown in ►**Fig. 5**.

Complications and Reoperations

Complication and reoperation data are compiled in ►**Table 2**. One recurrence case occurred in this period (2%). Another case (2%) presented an intraoperative L5 root lesion. Lumbar arthrodesis was required in 2 cases (4%; 1 case at the L4-L5 lumbar level, and 1 case at the L5-S1 lumbar level) 6 months postvideoendoscopy, due to a previous instability with discal decay and, consequently, to a foraminal stenosis with sciatica. There were no cases of postoperative deep or superficial infection.

Discussion

Lumbar disc herniation generating pain that irradiates to the lower limbs and is refractory to conservative treatment requires removal by the decompression of the neural structures, either of the isolated nerve root or of the dural sac.[16] Disc herniation surgery evolved from open discectomy procedures to microdiscectomy, and then to videoendoscopic discectomy. This evolution aims to reduce the surgical aggression to the surrounding tissues, but without altering the surgical objective, which is the release of nervous compression. When the resection of the surrounding structures is avoided or decreased, the less traumatic disc removal can reduce the collateral damage caused by the surgery.[17–19]

The full-videoendoscopic surgery is performed by a single portal, using an endoscope with an intraendoscopic working channel.[12] The technique has been successfully used, achieving the same goals of microdiscectomy, but also reducing perioperative local pain and surgical complications.[20–23] The transforaminal or the posterolateral routes are widely employed, but they present technical limitations, such as in cases with fragments located in the vertebral canal.[24] Thus, the interlaminar technique was developed to allow the removal of hernias that are not accessible through the transforaminal technique.[11,12] The present study is, in the knowledge of the authors, the first work in Portuguese to describe the results of this surgical option.

The final clinical results of the full-videoendoscopic procedure and of the surgical microdiscectomy have been very similar in some clinical trials regarding the reoperation index and the clinical improvement.[25,26] However, the advantages of the full-videoendoscopic surgery are highlighted. The adoption of these techniques improved low back pain, post-surgical rehabilitation, intraoperative complications, and tissue trauma.[25] The main implication is the possibility of carrying out the procedure with safety, even in patients with comorbidities and who demand a quick return to daily activities, because, compared with microdiscectomy, the incision is smaller, the blood loss is negligible, there is less damage to the paraspinal musculature, and the patient may be discharged earlier and already walking. For the surgeon, access is easy in obese and morbidly obese patients, and the visualization of structures is better due to the absence of blood and to the closer proximity using optics with saline irrigation.

Comparative clinical trials showed that microdiscectomy and the full-videoendoscopic technique yield very similar results regarding medium- and long-term resolution of the pain caused by neural compression,[25,27,28] with an improvement ranging from 75 to 90% in VAS scores, and from 67 to

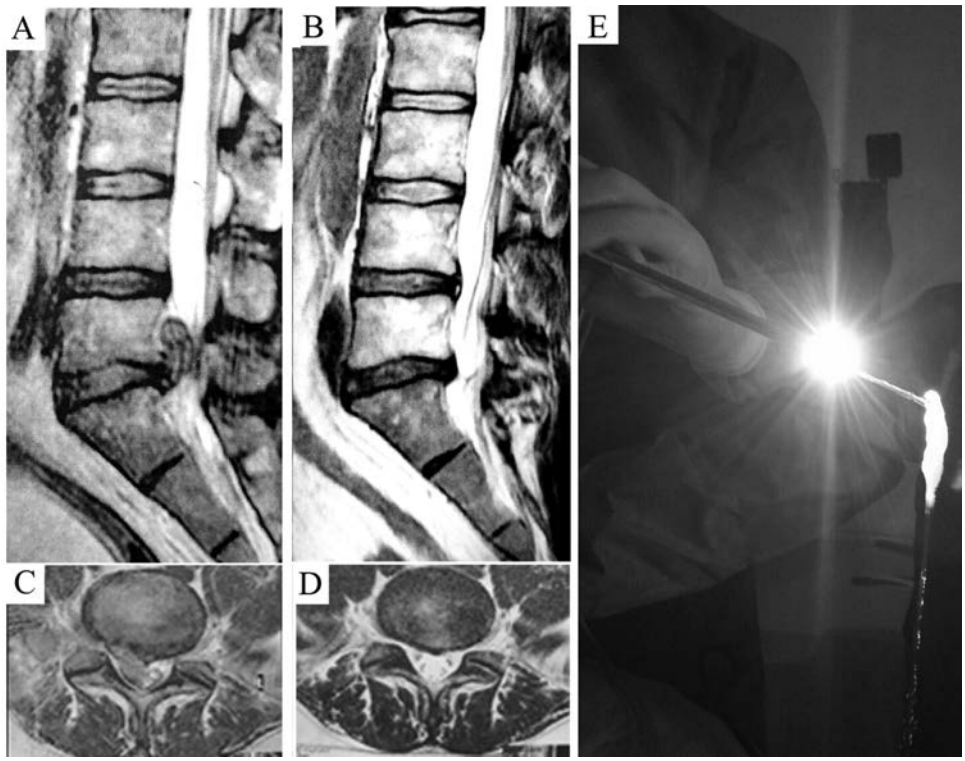

Fig. 5 Magnetic resonance images of the lumbar spine during the preoperative period in (A) sagittal section and in (B) axial section, and in the postoperative period in (C) sagittal section and in (D) axial section. Note the presence of a bulky fragment in the preoperative images, which was removed during the surgery (E), and the resulting decompression of the vertebral canal and the maintenance of bone and muscular integrity in the postoperative images.

80% in physical disability measured by the ODI. In the present case series, we have observed an improvement of 88% in VAS pain scores, and an 87% decrease in the physical restriction determined by the ODI questionnaire.

Consistently, an analysis of $>$ 100 thousand surgeries confirmed that less invasive approaches are associated with lower infection rates after lumbar discectomy.[29] The general intraoperative complication rate in less invasive discectomy is very low, totaling $\sim$ 1.6%, and it includes infection, re-herniation, neurological deficit, incisional hematoma, cerebrospinal fluid (CSF) fistula, pulmonary embolism, and acute myocardial infarction.[30] Remnant fragments are the main cause of procedural failure. Recurrence is a considerable event in these procedures, with results ranging from 4.2 to 12.5%, with an average value of 6% (2% in the present study), depending on the duration of the follow-up in each analysis.[25,27,28,31] Moreover, there is no difference in the hernia recurrence rate between microdiscectomy and the full-videoendoscopic technique. No durotomy was observed in our case series, which is in line with the literature, in which the average rate of non-intentional intraoperative durotomy ranges from 0 to 3%, with no statistical difference between the 2 techniques, which is also explained by its low incidence.[25,27,28] Severe complications were noted in 4 of the 50 cases in the present study (1 intraoperative neural injury, 1 recurrence, and 2 evolutions for an indication for arthrodesis). It was previously observed that non-severe complications are significantly higher in groups treated with microdiscectomy than in those submitted to the full-videoendoscopic technique.[17–19]

The main difference in complications among the two techniques is the lowest possibility of infection with the endoscopy, since it is performed with continuous irrigation. In the present study, there was no superficial or deep infection. The studied group routinely uses an extended 7-day therapeutic protocol with cephalexin, which is unusual, but cited in other studies, in order to reduce the incidence of infection (1.7% versus 4.3%).[32] However, this subject still requires larger, controlled studies comparing different protocols.[33] Despite heterogeneous reports among papers, the hospitalization time for microdiscectomy ranges from 1 to 7 days,[16,27,28,31,34,35] exceeding the time for less invasive surgeries, such as sequestrectomy and full-videoendoscopic discectomy, which can be performed in an outpatient setting,[13,27,36] without overnight stay and bed occupancy.

In summary, a meta-analysis on microdiscectomy and on the full-videoendoscopic technique shows that both options are very efficient for the surgical objective, but none has a broad superiority regarding clinical outcomes, complications or reoperations. However, the full-videoendoscopic technique has a shorter hospitalization time (outpatient procedure) and a shorter operative time.[37] Thus, it should be noted that a reduced hospitalization time means, in addition to less discomfort and greater satisfaction for the patient, a reduction of costs and demands to the health system, bringing

benefits to the society, as has already been observed in cost-benefit studies performed in other countries.[38–41]

Conclusion

The full-endoscopic surgery for lumbar disc herniation involving the spinal canal led to a satisfactory clinical resolution with a low complication rate; moreover, it was deemed feasible for an outpatient model without prolonged hospitalization.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Trigeminal Neuralgia Caused by Cerebellopontine Angle Tumors: Surgical Series

Neuralgia trigeminal causada por tumor do ângulo pontocerebelar: série de casos cirúrgicos

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Abstract

Introduction Cerebellopontine angle (CPA) tumors represent an important cause of persistent and refractory trigeminal neuralgia (TN). It is believed that ~ between 1 and 9.9% of the cases of patients presenting with TN painful manifestation are caused by space-occupying lesions.

Objective The objective of the present study is to describe the clinical and surgical experience of the operative management of patients presenting with secondary type TN associated with CPA tumors.

Method An observational investigation was conducted with data collection from patients with secondary type TN associated with CPA tumors who were treated with surgical resection of the space-occupying lesion and decompression of the trigeminal nerve from January 2013 to November 2016 in 2 different centers in the western region of the state of São Paulo, Brazil.

Results We operated on 11 consecutive cases in which TN was associated with CPA during the period of analysis. Seven (63.6%) patients were female, and 4 (36.4%) were male. Seven (63.6%) patients presented with right-side symptoms, and 4 (36.4%) presented with left-side symptoms. After 2 years of follow-up, we observed that 8 (72.7%) patients showed a complete improvement of the symptoms, with an excellent outcome, and that 3 (27.3%) patients showed an incomplete improvement, with a good outcome. No patient reported partial improvement or poor outcome after the follow-up. There was no operative mortality.

Conclusion Cerebellopontine angle tumors represent an important cause of TN and must be included in the differential diagnosis of patients presenting with refractory and persistent symptoms. Surgical treatment with total resection of the expansive lesion and effective decompression of the trigeminal nerve are essential steps to control the symptoms.

Keywords
- trigeminal neuralgia
- cerebellopontine angle tumors

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Resumo

Introdução Os tumores de ângulo pontocerebelar (APC) representam uma importante causa de neuralgia do nervo trigêmeo (NT) persistente e refratária. Cerca de 1 a 9,9% dos casos de pacientes com manifestação dolorosa do trigêmeo são causados por lesões expansivas.

Objetivo O objetivo do presente estudo é descrever a experiência clínica e cirúrgica do tratamento de pacientes que apresentam tipo secundário de NT associada a tumores de APC.

Método Foi realizada uma investigação observacional com coleta de dados de pacientes com NT secundária associada a tumor de APC tratados com ressecção cirúrgica de lesão expansiva e descompressão do nervo trigêmeo entre janeiro de 2013 e novembro de 2016, em 2 diferentes centros da região oeste do estado de São Paulo, Brasil.

Resultados Foram abordados 11 casos consecutivos nos quais NT foi associada a tumores de APC durante o período de análise. Sete (63,6%) pacientes eram do sexo feminino, e 4 (36,4%) do sexo masculino. Sete (63,6%) pacientes apresentaram sintomas no lado direito, e 4 (36,4%) apresentaram sintomas no lado esquerdo. Após 2 anos de seguimento, 8 (72,7%) pacientes apresentaram melhora completa dos sintomas com excelente resultado, e 3 (27,3%) pacientes apresentaram melhora incompleta com boa evolução. Não houve mortalidade operatória.

Conclusão Os tumores de APC representam uma importante causa de NT e devem ser incluídos no diagnóstico diferencial de pacientes com sintomas refratários e persistentes. O tratamento cirúrgico com ressecção total da lesão expansiva e descompressão efetiva do nervo trigêmeo são etapas essenciais para o controle dos sintomas.

Palavras-chave

- neuralgia trigeminal
- tumor do ângulo pontocerebelar

Introduction

Although the classical type of trigeminal neuralgia (TN) is the most common symptom of neurovascular conflict, some patients have the secondary type of TN, in which space-occupying lesions are responsible for the symptoms.[1–3] It is believed that ~ between 1 and 9.9% of the cases of patients presenting with TN painful manifestation are caused by cerebellopontine angle (CPA) tumors.[4] Meningiomas, schwannomas, and hemangioblastomas are extremely common posterior fossa tumors that may cause TN by directly compressing the trigeminal nerve or by being close to vascular structures around the nerve entry zone.[5–7]

The aim of the present study is to describe the clinical and surgical experience of the operative management of patients presenting with the secondary type of TN associated with CPA tumors.

Method

Delineation of the Study

An observational investigation was conducted with data collection from patients with the secondary type of TN associated with CPA tumors who were treated with surgical resection of the space-occupying lesion and decompression of the trigeminal nerve from January 2013 to November 2016 in 2 different centers in the western region of the state of São Paulo, Brazil. Clinical data were obtained retrospectively from the records and files of the patients. For all of the patients with the diagnosis of TN and radiological evidence of CPA tumor on magnetic resonance imaging (MRI), the following data were collected: gender, age at surgery, side of pain, type of the conflict/compression, type and number of medications used.

Presurgical Evaluation

An MRI of the brain was obtained from all of the patients with TN associated with CPA according to a specific protocol using a 1.5 Tesla MRI Scanner (Philips, Amsterdam, Netherlands) at the department of neuroradiology at our institution. All of the MRIs were analyzed by an experienced neuroradiologist who confirmed the visual radiological diagnosis of space-occupying lesion. High-resolution MRIs were performed to view the CPA anatomy of the patients and to exclude the presence of any additional neurovascular conflict.

Surgical Technique

The surgical approach was similar for all of the patients, and all of the procedures were performed by neurosurgeons experienced in microvascular decompression (MVD) surgery for TN. The surgery was performed under general anesthesia with a flexible spiral tracheal tube to allow the flexion of the neck while securing the airways. All of the patients were placed in the lateral position with the head supported with a three-pin Mayfield head fixation. A 5- to 7-cm curvilinear incision was made obliquely inside the hairline at the upper retromastoid area. A 1.5- to 2-cm diameter keyhole bone opening or small craniectomy or craniotomy was performed using a 4- and 4-mm extra-coarse power Diamond drill

system, EM 100-A Midas Rex Legend EHS, manufactured by Medtronic Powered Surgical Solutions - Menphis, TN, USA. The keyhole was located at the inner corner of the transverse sinus and the sigmoid sinus. Before the dural opening, precise hemostasis was accomplished with bone wax, Surgicel (Ethicon Inc., Bridgewater, NJ, USA) and cautery. The mastoid air cells were sealed with bone wax. The dura was opened in an inverted-T fashion, and small dural flaps were stitched to make a maximal dural opening (5 to 10 mm). The cerebrospinal fluid (CSF) was gradually aspirated and, under the operating microscope, an infratentorial lateral supracerebellar dissection was advanced to expose the petrosal vein (one to three bridging veins). Sufficient arachnoid dissection was performed around the petrosal veins and caudally to carefully expose the facial nerve and any offending vessels around the neural structures from proximal to distal. The space-occupying lesions were easily identified in all of the cases, and a circumferential dissection was initially performed whenever possible in order to carefully identify all of the neural and vascular structures before the debulking/resection of the tumor. Any additional compressing arterial loops or venous contacts were carefully dissected and mobilized off of the nerve root. A nonabsorbable material was interposed between the any vessel and the entry zone of the trigeminal nerve. Cerebellar retraction was judiciously used when necessary. A meticulous hemostasis and cleanup of the operating field was often achieved. Watertight dural closure with or without fascial graft was performed, and the cranioplasty was completed whenever possible. Cutaneous layers were routinely closed.

Outcome Assessment and Follow-Up

A 2-year follow-up investigation was performed in all of the patients included in the present study. The patients were clinically reassessed at 6, 12 and 24 months after the surgery, regarding the subjective improvement in the preoperative facial pain. They were included in 3 different groups according to the percentage of amelioration of pain, as follows: excellent—complete improvement ($>$ 90%); good—incomplete improvement (75–90%); and poor – partial improvement ($<$ 75%).

Ethical Statement

The ethical committee of our institution analyzed the project and approved the performance of our investigations. All of the patients have given their informed consent for participation in the research study. The present study complied with the Declarations of Helsinki and Nuremberg. Informed consent for surgery was acquired from all of the patients.

Statistical Analysis

The data collected from all of the patients were organized in tables. The data are expressed as the means $\pm$ the standard deviation (SD) for parametric variables, and as the median values for nonparametric variables. A normal distribution to sample collected data was assumed. The statistical analyses and the review of the numerical results obtained in the present investigation were performed by a mathematical team, and a p-value < 0.05 was considered statistically significant.

Results

We have operated on 11 consecutive cases in which TN was associated with a CPA tumor during the period of analysis. Seven (63.6%) patients were female, and 4 (36.4%) were male. Age distribution and duration of the symptoms are presented in ►**Table 1**. Seven (63.6%) patients presented with right-side symptoms, and 4 (36.4%) patients presented with left-side symptoms. After 2 years of follow-up, we observed that 8 (72.7%) patients showed a complete improvement of the symptoms, with an excellent outcome, and that 3 (27.3%) patients showed an incomplete improvement, with a good outcome. No patient reported partial improvement or poor outcome after the follow-up. There was no operative mortality. One (9.09%) patient presented with CSF leak, which was surgically treated on the 7th postoperative day, and 2 (18.2%) patients presented with temporary facial palsy.

Table 1 Clinical details of patients with trigeminal neuralgia and cerebellopontine angle tumors

Patient	Age (years old)	Gender	Duration of the symptoms (months)	Side	Outcome	Complication	Tumor
1	63	Female	24	Right	Excellent	–	Meningioma
2	59	Female	36	Right	Good	–	Meningioma
3	67	Female	18	Left	Excellent	CSF leak	Meningioma
4	61	Male	33	Right	Excellent	–	Meningioma
5	64	Female	27	Right	Excellent	–	Meningioma
6	52	Male	31	Left	Good	Facial palsy	Schwannoma
7	47	Male	19	Left	Excellent	–	Schwannoma
8	61	Female	42	Right	Good	Facial palsy	Schwannoma
9	49	Male	18	Left	Excellent	–	Schwannoma
10	33	Female	14	Right	Excellent	–	Hemangioblastoma
11	37	Female	16	Right	Excellent	–	Hemangioblastoma

Abbreviations: CSF, cerebrospinal fluid.

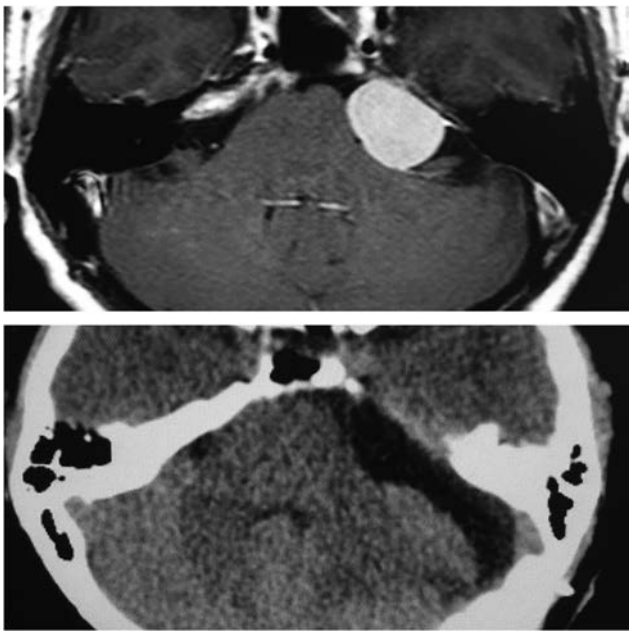

Fig. 1 Cerebellopontine angle tumor meningioma compressing the trigeminal nerve. Postoperative computed tomography.

Discussion

Several types of pathologies occur in the posterior fossa, and the specific diagnosis relies on the location of the lesion, on the age of the patient, on the associated clinical history, including signs, symptoms and duration, and on imaging studies.[8–11] Painful manifestation of posterior fossa tumors compressing the TN is a frequent manifestation of expansive lesions occupying the CPA.[9–12] It is believed that ~ between 1 and 9.9% of the cases of patients presenting with trigeminal painful manifestations are caused by CPA tumors, including meningiomas, schwannomas, and hemangioblastomas.[4–7] In the present study, we describe our clinical and surgical experience treating patients with TN secondary to posterior fossa tumors located at the CPA.

Vestibular schwannomas and meningiomas are the 2 most frequent lesions of the CPA and account for ~ between 85 and 90% of all of the posterior fossa fossa tumors.[5] ►**Figure 1** and **2** exemplifies CPA tumor meningioma and a large vestibular schwannoma, respectively. Liu et al (2017) found CPA meningiomas as the most common causes of tumor-induced TN, accounting for 45.7% of the patients with symptomatic TN in this study.[7] According to the authors, meningiomas tended to displace the nerve with or without vascular compression on

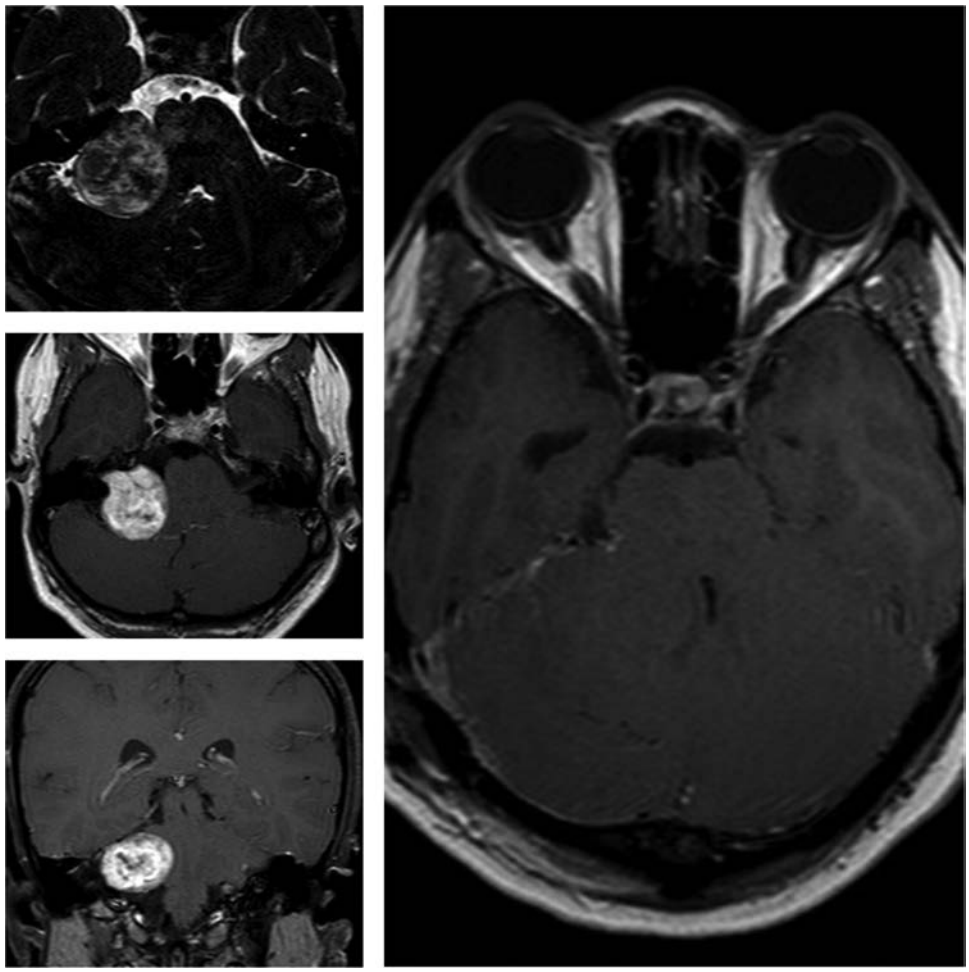

Fig. 2 Large vestibular schwannoma compressing the trigeminal nerve. Postoperative magnetic resonance imaging.

the entry zone.[7] In our investigation, meningiomas were also found as the most frequent posterior fossa tumors associated with TN, accounting for 45.5% of the patients with symptomatic TN. In accordance with previous publications, we have verified that tumor removal produces > 95% of symptom control in most patients.[7,13,14]

Large vestibular schwannomas are also described as potentially expansive lesions associated with TN that usually present no symptom improvement after stereotactic radiosurgery.[15] Neff et al (2017) showed that 16% of the patients with large vestibular schwannomas present with TN, and that the removal of the tumor alleviated pain in 70% of the cases.[15] Twenty-five percent of the patients did not show any change in the intensity of the neuralgia after the operation.[15] In the present study, we have observed that an excellent outcome regarding pain relief was achieved in 50% of the patients. Postoperative facial palsy was directly related with limitations in the control of the symptoms, as observed by others.[7,15]

Hemangioblastomas are tumors less frequently associated with TN.[7] They represent < 1% of the cases in some series.[7] However, tumor resection is usually associated with few complications and satisfactory symptom control.[7] In our investigation, we could operate on two young women with short symptom duration, resulting in an excellent control of the neuralgia after the surgery. Also, no complication related to the procedure was noted.

Complications associated with tumor removal to treat TN due to compressive symptoms are not rare. Liu et al (2017) described CSF leak, hearing disturbances, facial palsy, facial numbness, and oculomotor paralysis as possible complications when trying to decompress the trigeminal nerve from expansive lesions.[7] In the present study, we observed 1 (9.1%) case of CSF leak, which was treated with reoperation, and 2 (18.2%) cases of temporary facial palsy in large vestibular schwannomas. All of the patients with schwannomas included in the present investigation were preoperatively deaf, and their hearing status did not change after the surgery. There was no operative mortality.

Conclusion

Cerebellopontine angle tumors represent an important cause of TN and must be included in the differential diagnosis of patients presenting refractory and persistent symptoms. Surgical treatment with total resection of the expansive lesion and effective decompression of the trigeminal nerve are essential steps to control the symptoms.

Conflicts of Interest
The authors have no conflicts of interest to declare.

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Effect of Albendazole Treatment in an Experimental Model of Neurocysticercosis-Induced Hydrocephalus

Efeito do tratamento com albendazol num modelo experimental de hidrocefalia induzida por neurocisticercose

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Abstract

Hydrocephalus is a major concern in neurocysticercosis (NCC), and its management is more challenging than that of hydrocephalus caused by other etiologies. Even though albendazole is a well-established drug for the treatment of NCC, the death of the parasites may worsen the clinical symptoms and eventually, deteriorate the course of hydrocephalus. The aim of this study was to analyze the effects of treatment with albendazole on the course of hydrocephalus as well as on animal behavior in a rat model of NCC-induced hydrocephalus in order to verify whether the course of hydrocephalus and the animal behavior were changed. Ventricle volumes before and after treatment showed a slight but non-statistically significant difference (168.11 mm^3 versus 184.98 mm^3, $p = 0.45$). The distribution and location of the cysts were unaffected. In addition, the behavioral patterns before and after the treatment were not significantly different, as assessed by the open field test. On histologic assessment, mononuclear leukocyte infiltration was present in diverse sites, such as the perivascular and peri-ependymal regions, choroid plexus, and meningeal membranes. A positive correlation was found between the degree of ventricle enlargement and tissue damage. Further studies with long-term comparisons are required.

Keywords
- hydrocephalus
- neurocysticercosis
- albendazole
- experimental design

Resumo

A hidrocefalia é uma das principais complicações da neurocisticercose (NCC), e seu manejo pode ser mais desafiador do que outras forma de hidrocefalia. Apesar de o albendazol ser uma droga bem estabelecida para o tratamento da NCC, a morte do parasitas pode agravar os sintomas clínicos e, eventualmente, deteriorar o curso de hidrocefalia. O objetivo deste estudo foi analisar os efeitos do tratamento com albendazol no curso da hidrocefalia, bem como no comportamento animal em um

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modelo de hidrocefalia induzida por NCC em ratos. Na análise do volumes ventricular antes e depois do tratamento, houve uma diferença leve, mas não estatisticamente significativa (168,11 mm^3 versus 184,98 mm^3, $p = 0,45$). A distribuição e localização dos cistos não foram afetadas.

Além disso, os padrões comportamentais antes e após o tratamento também não foram significativamente diferentes. A avaliação histológica mostrou infiltração leucocitária mononuclear em diversos sítios, como nas regiões perivascular e periependimária, plexo coróide e meninges. Uma correlação positiva foi encontrada entre o grau de aumento do ventrículo e dano tecidual. Mais estudos com comparações de longo prazo são necessários para melhor compreensão da possível deterioração clínica da hidrocefalia por NCC quando do tratamento clínico.

Palavras-chave
- hidrocefalia
- neurocisticercose
- albendazol
- desenho experimental

Introduction

Neurocysticercosis (NCC) is the most common parasitic disease of the central nervous system (CNS) worldwide.[1,2] Although it is potentially eradicable, it remains endemic in developing countries, and migratory flows have reintroduced the disease in Europe and the USA.[3,4]

Neurocysticercosis is a pleomorphic disease because *Taenia solium* cysts may lodge anywhere in the CNS and the parasite-host interaction is highly heterogeneous.[5] The extraparenchymal form of NCC has a more aggressive course because cysts in the cerebrospinal fluid (CSF) compartments can elicit an intense inflammatory reaction, leading to vasculitis, hydrocephalus, and increased intracranial pressure.[6,7]

Hydrocephalus is a major concern in extraparenchymal NCC, as patients with NCC-induced hydrocephalus have higher rates of morbidity and mortality. The management of NCC-related hydrocephalus is also more challenging than that of hydrocephalus of other etiologies because of the higher rates of shunt malfunction, infection, and the need for reoperation.[8–11]

Albendazole is a well-established drug for the treatment of neurocysticercosis. Although some controversy regarding the safety of albendazole in the treatment of extraparenchymal NCC has arisen in the last decades, albendazole remains the drug of choice even for giant extraparenchymal cysts with a mass effect. However, caution in the use of albendazole is needed because the drug accelerates the death of the parasites in the CSF compartments, worsening the clinical symptoms and eventually deteriorating the course of hydrocephalus.[12,13]

The pathophysiologic mechanisms of NCC-induced hydrocephalus are not fully understood, although experimental models have helped solve some questions regarding leukocyte infiltration and inflammatory reactions—mainly for the parenchymal form of the disease.[14–19] Recently, we developed an experimental model of NCC-induced hydrocephalus that reproduces magnetic resonance imaging (MRI) and histologic findings of human NCC.[20] In the present study, we aimed to analyze the effects of albendazole treatment in a rat model of NCC with concomitant hydrocephalus to evaluate the correlation between the course of hydrocephalus and animal behavior.

Methods

Animals

The Institutional Animal Care and Use Committee analyzed and approved the project. The animals were handled according to ethical guidelines and current legislation. Eighteen adult Wistar rats weighing $\sim$ 150 g were used. The rats were kept in rooms with controlled humidity and temperature (21°C) under a regular light cycle of 12/12 hours. Food and water were available *ad libitum*.

Sample size

According to previous studies, five animals are sufficient to determine statistical differences between groups in ventricle volume. Assuming a mortality rate of 30% immediately after the inoculation procedure and during the observational period, we used 9 animals for each group (18 in total). This sample size was estimated assuming a random sample with type I and II errors of 0.05 and 0.02, respectively, and a normal distribution of ventricle volumes without potential confounding factors.

Experimental Design

Eighteen rats weighing **150–200 g** were inoculated with 50 cysts of *Taenia crassiceps* (a *T. solium* analog cestode). Two animals died immediately after the inoculation. Thus, 16 rats were kept for observation. Three months after the inoculation, the rats underwent MRI and the open field test (OFT) to evaluate behavior patterns. The rats were then randomly divided into two groups: treatment with albendazole and no treatment (control group). One week after the treatment, the animals again underwent MRI and the OFT, and were euthanized for histologic assessment. ►**Fig. 1** shows a flowchart of the experimental design.

Parasites and Inoculation

The methods for the maintenance and inoculation of *T. crassiceps* have been described previously.[21] Briefly, after general intraperitoneal anesthesia with ketamine and

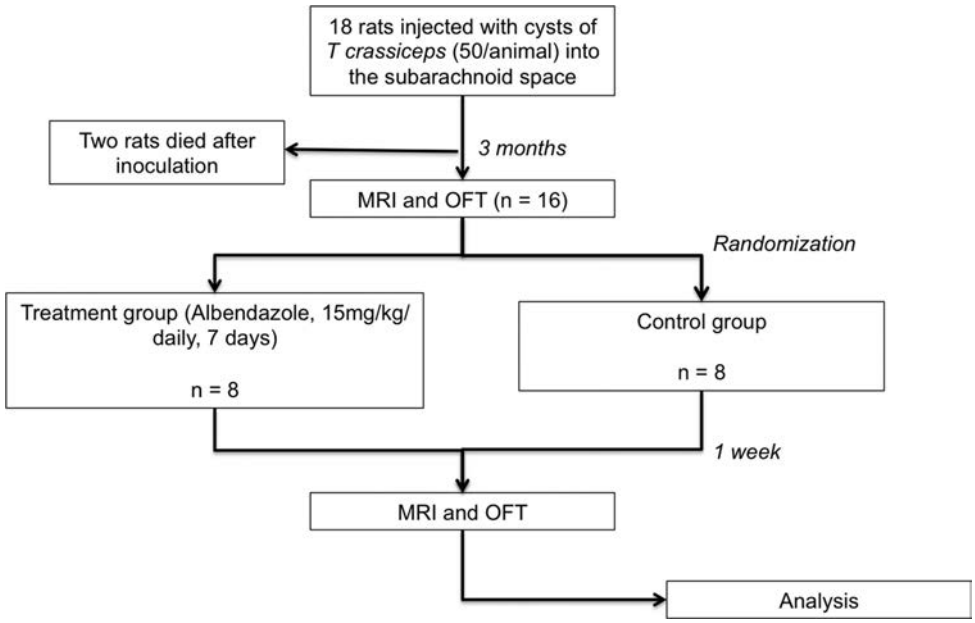

Fig. 1 Flowchart of the experimental design.

xylazine, the rats were inoculated with 50 metacestodes of *T. crassiceps,* each one, removed from the peritoneal cavities of infected mice, into the subarachnoid space (cisterna magna).

MRI

An MRI was performed using a 0.25 Tesla equipment, model Vet-MR (Esaote, Genoa, Italy). Improved visualization of ventricles and cysts was achieved using a T2-weighted gradient-echo sequence (0.6 mm thickness; T eco 5 ms; T repetition 10 ms). An experienced team analyzed the MRI findings focusing on cyst distribution. Volumetric analysis was performed using the ITK-SNAP software, version 3.0.0 (Penn Image Computing and Science Laboratory, Pennsylvania, USA).

Open Field Test

We used a wood chamber of 72 × 72 cm gridded with 9 equally sized squares (18 × 18 cm). Rats were placed in the chamber, and their behavior was recorded using a GoPro digital camera for 5 minutes. The records were analyzed by two independent observers registering the number of crossed lines, center time, stretch attend posture, and defecation. When the disagreement between the observers was lower than 10%, we used the higher value. When the disagreement was higher than 10%, the observers reexamined the record together to determine a final consensus value.

Histologic Assessment

After the second OFT, the animals were euthanized with an overdose of pentobarbital for encephalon removal. We used heart perfusion with, and overnight immersion in, 10% neutral-buffered formalin for brain fixation. The next day, slices were cut at the level of the optic chiasm, dehydrated in increasing concentrations of alcohol, diafanized in xylene, and embedded in paraffin. The paraffin blocks were cut into 3-µm sections, and the sections were stained with hematoxylin-eosin. For histological assessment, we used the criteria of Matos-Silva for experimental encephalitis, focusing on meningeal, perivascular, ependymal, and choroid plexus leukocyte infiltration, periventricular gliosis and edema, choroid plexus edema, ependymitis and ependymal destruction and hyperemia. These parameters were graded semi-quantitatively (absent: 0 points, light: 1 point, moderate: 2 points, and severe: 3 points). The total score of tissue damage was the sum of the scores of each parameter.

Statistical Analysis

The BioEstat 5.3 software (BioEstat Software, Belém, PA, Brazil) was used to assess the normal distribution of variables (Shapiro-Wilk test), differences between pre and post-treatment ventricle size (t-test), correlation between OFT results and ventricle size (Pearson correlation), and correlation between histologic grades and ventricle size (Spearman correlation). Statistical significance was set at $p = 0.05$.

Results

Of the 16 inoculated rats, nine developed hydrocephalus (the cutoff value for a normal ventricle volume was 5.0 mm^3). ►**Fig. 2** shows an example of a hydrocephalic animal with cysts in the CSF compartments. Eight of these nine hydrocephalic animals were in the treatment group at randomization. Because the groups were not comparable, we excluded the control group from the ventricle and behavioral analyses, and focused our assessment on pre- and post- treatment differences.

Ventricle volumes before and after treatment showed a slight but non-significant difference (168.11 mm^3 versus 184.98 mm^3, $p = 0.45$). The distribution and location of cysts did not change, except in one animal that had no cysts within

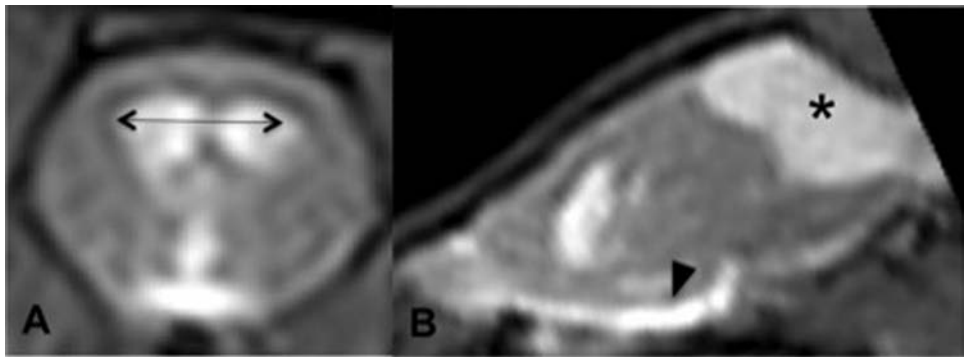

Fig. 2 Magnetic resonance imaging showed hydrocephalus with ventricular enlargement (**A**, arrow) and cysts in the basal cistern (B, arrowhead) and in the cisterna magna (**B**, asterisk) in a *T. crassiceps*-inoculated mouse.

the basal cisterns pre-treatment but showed cysts after treatment. The results are summarized in ►**Table 1**.

Regarding the behavioral pattern in the OFT, we found no differences before and after treatment (line crossing: $p = 0.73$; center time: $p = 0.35$; stretch attend posture: $p = 0.73$; defecation: $p = 0.45$). In addition, no correlation was found between the ventricle volume and OFT pattern variations ($p > 0.05$).

On histologic assessment, we were able to verify the occurrence of mononuclear leukocyte infiltration in diverse sites, such as the perivascular and peri-ependymal region, choroid plexus, and meningeal membranes (►**Fig. 3**). To determine whether ventricle volume was associated with the histologic damage score, we considered all 16 animals and found a positive correlation (rho coefficient = 0.53; $p = 0.04$).

Discussion

Neurocysticercosis is a pleomorphic disease, and the full understanding of its pathophysiologic mechanisms is hindered by the heterogeneity of the parasite-host interaction and by the interference of drugs commonly employed to treat NCC in clinical practice, such as cysticides and corticosteroids.[22] For this reason, many investigators have attempted to establish experimental models of the disease.[23]

The use of albendazole for extraparenchymal NCC remained controversial for some time; however, it is currently well-accepted and recommended.[24] Nevertheless, the risk of exacerbation of symptoms, notably deterioration of hydrocephalus, should be always borne in mind when prescribing albendazole.[25,26] We aimed to examine whether the use of albendazole in an experimental model of extraparenchymal NCC could reproduce this potential adverse effect.

We found that neither the hydrocephalus nor behavioral patterns changed significantly in the short term after the albendazole treatment. However, we cannot state that albendazole was a safe drug in our experimental model since the drug was not effective either, as the cysts remained visible in the CSF spaces in the MRI analyses. Thus, the drug was not sufficiently effective to completely eliminate the parasite in the short term.

Similarly, in clinical practice, the analysis of a subgroup from a large randomized controlled trial did not show increased rates of disappearance of extraparenchymal cysts in patients even 12 months after albendazole treatment.[27] In fact, few controlled trials of medical treatment for

Table 1 Ventricle volume of each animal in the treatment group before and after the treatment, and the distribution of cysts in the main cerebrospinal fluid (CSF) compartments

Animal	Pretreatment					Posttreatment				
	Ventricle volume (mm^3) *	Cisterna magna	Brain convexity	Basal cisterns	Intra-ventricular	Ventricle volume (mm^3) *	Cisterna magna	Brain convexity	Basal cisterns	Intra-ventricular
1	107.20	Yes	No	Yes	No	26.25	Yes	No	Yes	No
2	46.35	Yes	Yes	No	No	43.53	Yes	Yes	Yes	Yes
3	291.70	Yes	No	Yes	Yes	324.0	Yes	No	Yes	Yes
4	68.08	No	No	Yes	No	88.17	No	No	Yes	No
5	177.10	No	No	No	No	165.4	No	No	No	No
6	17.80	No	No	Yes	No	29.81	No	No	Yes	No
7	69.34	Yes	No	Yes	No	101.20	Yes	No	Yes	No
8	567.30	Yes	No	Yes	No	701.50	Yes	No	Yes	No

*The mean ventricle volume before and after treatment was 168.11vmm^3 (±183.46) and 184.98vmm^3 (±230.58), respectively. This difference did not reach statistical significance ($p = 0.45$). Only animal #2 showed a difference in the presence of cysts in the basal cisterns pre and posttreatment.

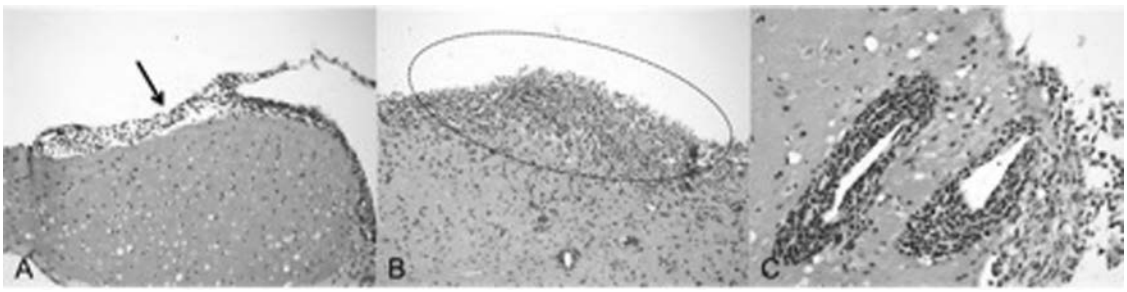

Fig. 3 Histologic assessment showed meningeal mononuclear leukocyte infiltration (**A**, arrow), a pattern of chronic ependymitis (**B**, dotted lines), and intense perivascular leukocyte mononuclear infiltration (**C**).

extraparenchymal NCC have been published, and most of the current knowledge is based on case series.[28] Experimental models can contribute to filling this evidence gap.

In addition, albendazole dosage should also be considered. Góngora-Rivera et al (2006) proposed that a higher dose (30 mg/kg/day) would be more effective for larger cysts in subarachnoid and intraventricular cysticercosis.[29] The concomitant use of steroids is also recommended to reduce inflammatory reactions.[30] To date, we have observed inflammatory cells in different analyzed brain regions, and we believe that future experiments using steroids may be useful to demonstrate the reduction of inflammation.

Finally, the present study adds new information on the experimental model of NCC-induced hydrocephalus. We have previously observed inflammation, edema, gliosis, and ependymal destruction in this model. In the present study, we were able to demonstrate that the degree of hydrocephalus was associated with the severity of tissue lesions.

Some limitations must be pointed out. First, we observed high heterogeneity of the disease between the control and treatment groups. Future studies should only use animals with verified hydrocephalus for randomization. Second, it is not clear whether the parasite mass-effect or the inflammation was more important in the development of hydrocephalus. Demonstrating that hydrocephalus can occur even with the parasites destroyed will guide the understanding of the role of inflammation in this model of extraparenchymal NCC.

Conclusion

In the short term, albendazole did not deteriorate the course of hydrocephalus and behavioral patterns in a model of neurocysticercosis-induced hydrocephalus.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Radiation Exposure during Percutaneous Endoscopic Lumbar Discectomy: Interlaminar versus Transforaminal

Exposição à radiação durante discectomia endoscópica lombar percutânea: interlaminar versus transforaminal

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Abstract

Objective Percutaneous endoscopic lumbar discectomy (PELD) relies heavily on fluoroscopy guidance; therefore, medical staff exposure to radiation has become an important issue. The purpose of this study was to determine the radiation dose and the amount of time to which the surgeons are exposed during PELD and to compare both parameters in the transforaminal (TF) and interlaminar (IL) approaches. Although they are considerably different, they may be wrongly considered together.

Methods A retrospective evaluation of the last 20 PELD performed by the authors is presented. Patients were distributed in 2 groups. Six (1F, 5M) patients were submitted to IL-PELD and 14 (6F, 8M) to TF-PELD. Fluoroscopy reports were obtained from patients' records, all performed with the same C-Arm device and software mode. Groups were compared using unpaired *t*-test.

Results The IL group showed an average radiation exposure of 8.37 $\pm$ 4.21 mGy and duration of 11.1 $\pm$ 5.45 seconds, while the TF group showed an average radiation exposure of 28.92 $\pm$ 7.56 mGy and duration of 42 $\pm$ 16.64 seconds. The *p*-value for radiation was 0.0000036, and for time it was 0.00027.

Conclusions Interlaminar PELD requires a lower radiation dose and a shorter amount of exposure than TF-PELD. Studies that concern radiation required for minimally-invasive spine surgeries should consider the PELD approaches separately.

Keywords
- intervertebral disc displacement
- fluoroscopy
- background radiation
- artroscopy

Resumo

Objetivo A discectomia endoscópica lombar percutânea (DELP) depende muito de orientação por fluoroscopia; portanto, a exposição à radiação se tornou um assunto importante. O objetivo deste estudo foi determinar a dose e o tempo de radiação aos quais os cirurgiões estão expostos durante a discectomia endoscópica lombar percutânea (DELP) e comparar ambos os parâmetros nos acessos transforaminal (TF) e interlaminar (IL). Embora sejam consideravelmente diferentes, estes podem ser erroneamente considerados em conjunto.

Métodos Avaliação retrospectiva dos últimos 20 casos de DELP realizados pelos autores. Os paciente foram distribuídos em dois grupos. Seis (1M, 5H) pacientes foram submetidos a DELP-IL e 14 (6M, 8H) a DELP-TF. Os dados da fluoroscopia foram obtidos

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dos relatórios dos pacientes, todos avaliados usando o mesmo aparelho de arco cirúrgico e no mesmo modo do programa. Os grupos foram comparados utilizando o teste-*t* não pareado.

Resultados O grupo IL mostrou exposição média à radiação de 8,37 ± 4,21 mGy e duração de 11,1 ± 5,45 segundos, enquanto o grupo TF apresentou exposição média à radiação de 28,92 ± 7,56 mGy e duração de 42 ± 16.64 segundos. O valor de *p* para a radiação foi de 0.0000036 e para o tempo foi de 0.00027.

Conclusões A DELP-IL necessita de menor quantidade de radiação e tempo do que a DELP-TF. Estudos cujo interesse é a radiação para cirurgias minimamente invasivas da coluna devem considerar os diferentes acessos para DELP separadamente.

Palavras-chave
- deslocamento do disco intervertebral
- fluoroscopia
- radiação de fundo
- artroscopia

Introduction

Lumbar discectomy has traditionally been performed through microdiscectomy, an open surgical technique. Refinement of operative techniques, a better understanding of the anatomy, and the development of novel technology has led to less invasive surgical options, such as percutaneous endoscopic lumbar discectomy (PELD). Several trials have demonstrated the effectiveness of PELD; however, minimally invasive spine surgeries (MISS) such as PELD rely heavily on intraoperative navigation, and fluoroscopy guidance is usually chosen for a safe percutaneous approach and accurate localization.[1–5] Therefore, medical staff exposure to radiation has become an important issue.[1,6–8] Some publications have addressed the amount of radiation exposure in various percutaneous spine procedures,[2,9–12] including PELD,[1] but none have compared the amount of radiation in transforaminal (TF) and interlaminar (IL) approaches. Although the approaches are considerably different, they may be wrongly considered together when the topics are radiation exposure and PELD.

This study was performed considering the hypothesis that the IL-PELD uses a considerably lower amount of radiation, and that this would constitute an advantage over the TF-PELD. The purpose of this study was to determine the radiation dose and the amount of time to which the surgeons are exposed during PELD using a particular C-Arm device (Ziehm Solo, Ziehm Imaging, Nürnberg, Germany) and to compare both parameters in the TF and IL approaches.

Methods

Surgical Technique Details

Transforaminal PELD was typically performed as described previously[1,13,14] and consisted of 2 parts: a TF approach under fluoroscopic control followed by selective discectomy with endoscopic visualization. The first part consists of insertion of the needle into the disc (►**Fig. 1A**), injection of contrast and methylene blue (►**Fig. 1B**), then, the needle is replaced with a guide wire, and an obturator is introduced along the guide wire (►**Fig. 1C**). After the obturator position is confirmed, a bevel-ended working sheath is placed near the disc-herniation (►**Fig. 1D**–**E**). For safe introduction of the spinal needle, obturator and working sheath through the foraminal window, a real-time anteroposterior (AP) and/or lateral view in the C-Arm device is essential. After insertion of the endoscope, the first part of the procedure is over, as well as the mainly use of fluoroscopy. Surgery is now performed under direct endoscopic visualization for discectomy using mainly forceps and radiofrequency coagulation. In this second part, fluoroscopy is used only seldom to check the position of the instruments and double check anatomic parameters (►**Fig. 1F**).

Interlaminar PELD also consisted of 2 parts. However, in this case, the first part, the fluoroscopic guided IL approach, is typically faster than the in the TF approach. While the TF approach is initiated with the insertion of a needle, in the IL approach, the obturator can be directly inserted after IL window identification in the AP view (►**Fig. 2A**) and a 1-cm skin incision, without the use of a needle. Lateral view is used to check the position and direction of the obturator, and the working sheath is then inserted, completing the first part of the procedure (►**Fig. 2B**). Surgery is then performed under direct endoscopic visualization for discectomy using mainly scissors, forceps, dissectors and radiofrequency coagulation. In this second part, fluoroscopy is used only seldom to check the position of the instruments and double check anatomic parameters (►**Fig. 2C**).

Study Design and Sample

The last 20 patients of this Institution that underwent PELD between May and December 2017, performed by the authors, were included in this retrospective study. Only those patients that underwent single-level, unilateral endoscopic decompression were included. No cases of decompressions adjacent to a fusion were included, and neither were those who needed foraminal or central stenosis treatment. Patients who did not have radiological data and archive were excluded. All data were acquired from the same C-Arm device (Ziehm Solo, Ziehm Imaging, Nürnberg, Germany) and performed in the same software mode. Surgeries performed with other devices or modes were not included. The surgeons had, at the time of the study, more than 6 years of endoscopic spine surgery experience.

Results

Six patients underwent IL-PELD, 1 female and 5 male, and 14 patients underwent TF-PELD, 6 female and 8 male (►**Table 1**).

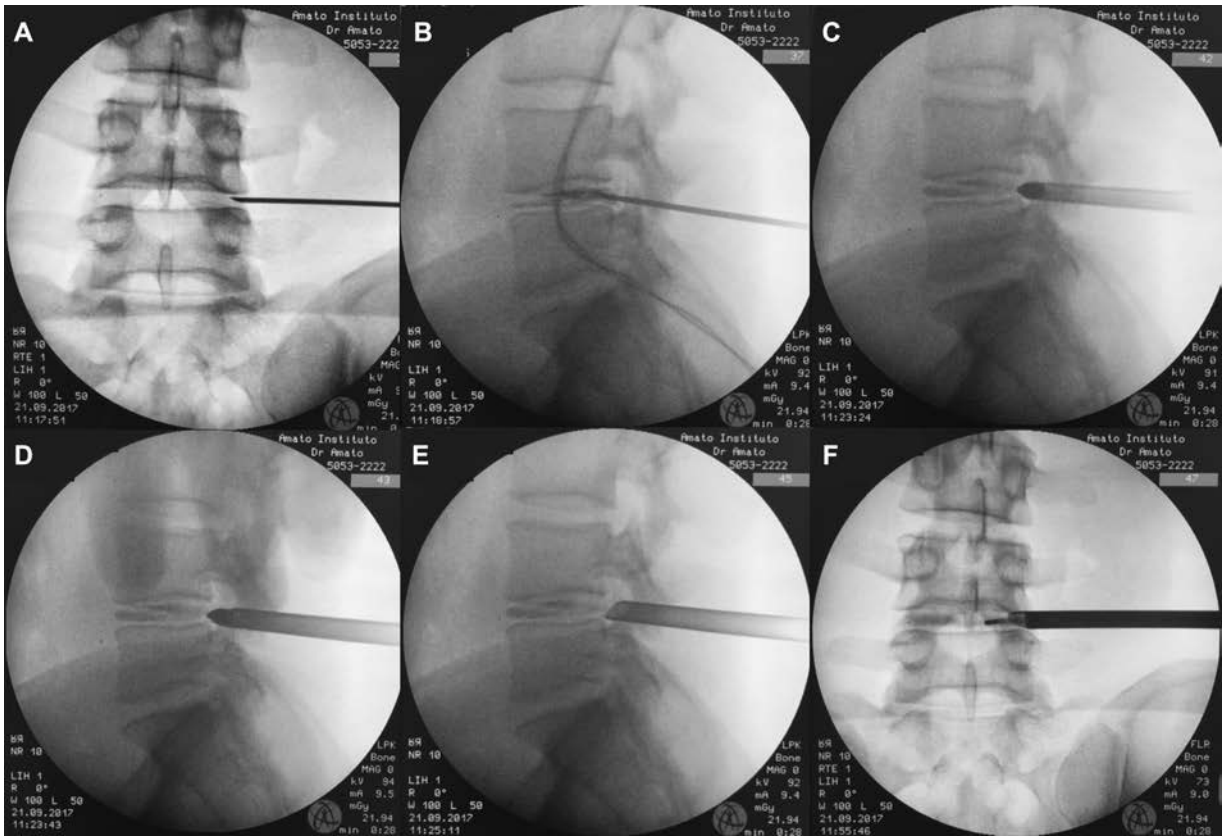

Fig. 1 Transforaminal percutaneous endoscopic discectomy. Fluoroscopic anteroposterior view (**A**) shows needle insertion into the disc. Lateral view (**B**) shows discography with contrast injection, then, obturator insertion (**C**), followed by working sheet insertion (**D**) and obturator withdraw. Anteroposterior view (**F**) shows working instrument inside the disc. Fluoroscopic data of patient number 6 are also displayed.

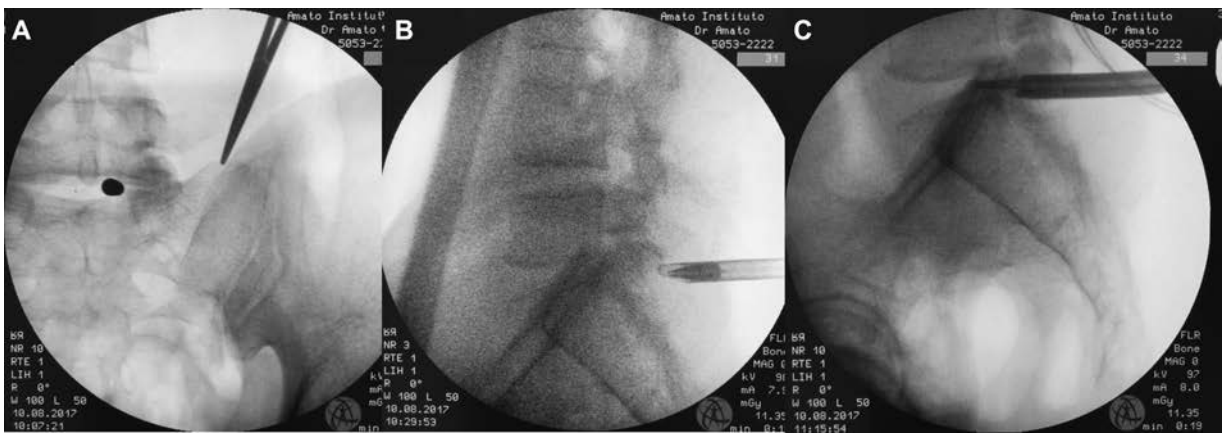

Fig. 2 Interlaminar percutaneous endoscopic discectomy. fluoroscopic anteroposterior view (**A**) shows direct insertion of the obturator. Lateral view (**B**) shows insertion of the working sheet. (**C**) shows working sheet and endoscope inside the spinal canal and a working instrument inside the disc. Fluoroscopic data of patient number 4 are also displayed.

The mean age of the patients was 46 years in IL-PELD and 46.8 years in IL-PELD. The average time of fluoroscopy was 11.2 ± 5.5 seconds in the IL group against 42.9 ± 16.6 in the TF group ($p = 0.00027$), and the amount of fluoroscopy exposure was $8.37 + 4.21$ mGy in the IL group against $28.92 + 7.56$ mGy in the TF group ($p = 0.0000036$) (►**Table 2**).

Discussion

Minimally invasive spine surgery and needle-based interventional spine procedures still typically depend on ionizing radiation for localization and guidance in placing the equipment,[15] although different strategies have been adopted to reduce medical staff and patient exposure to radiation as

Table 1 Individual characteristics and results of the 20 patients

N	Age	G	Approach	Level and characteristics	Time (sec)	Radiation (mGy)
1	55	F	TF	L3L4 - foraminal - extrusion	00:48	28.036
2	51	M	TF	L5S1 - foraminal - extrusion	00:40	29.350
3	71	M	TF	L4L5 - posterolateral - extrusion	00:45	35.296
4	35	M	IL	L5S1 - posterolateral - extrusion	00:19	11.352
5	40	F	TF	L1L2 - central - extrusion	00:59	40.704
6	30	M	TF	L4L5 - posterolateral - extrusion	00:28	21.939
7	63	M	TF	L4L5 - posterolateral - protrusion	00:44	37.110
8	54	M	TF	L4L5 - posterolateral - down migration	00:34	23.871
9	42	M	TF	L4L5 - posterolateral - down migration	00:23	15.254
10	41	M	IL	L5S1 - posterolateral - extrusion	00:09	4.236
11	59	M	TF	L4L5 - posterolateral - extrusion	00:34	17.308
12	41	F	TF	L5S1 - central - extrusion	00:50	25.510
13	41	F	TF	L4L5 - central - extrusion	01:30	30.400
14	38	M	IL	L4L5 - posterolateral - down migration	00:09	13.860
15	72	M	IL	L5S1 - central - extrusion	00:06	5.115
16	32	M	TF	L4L5 - posterolateral - extrusion	00:42	31.963
17	39	F	TF	L4L5 - posterolateral - extrusion	00:35	37.490
18	37	F	TF	L5S1 - foraminal - extrusion	00:29	30.620
19	49	F	IL	L5S1 - posterolateral - extrusion	00:17	11.079
20	41	M	IL	L5S1 - posterolateral - extrusion	00:07	4.566

Abbreviations: F, female; G, gender; IL, interlaminar; M, male; mGy, miligray; n, number; sec, seconds; TF, transforaminal.

Table 2 Statistical analysis

	Time - IL (sec)	Time - TF (sec)	Radiation - IL (mGy)	Radiation TF (mGy)
Average	11.2	42.9	8.37	28.92
SD	5.5	16.6	4.21	7.56
***p*-value**	0.0003		0.000004	

Abbreviations: IL, interlaminar; mGy, miligray; SD, standard deviation; sec, seconds; TF, transforaminal.

well as to reduce operating time, such as ultrasound-assisted TF-PELD[16] and preoperative location methods.[17]

It is known that among various discectomy techniques, the radiation dose is greater on PELD and other MISS when compared with open surgery.[1,2] That is understandable, since fluoroscopy is used to help on anatomical identification throughout the non-open surgeries. Mariscalco et al also show that tubular microdiscectomy uses smaller doses of radiation than TF-PELD.[2] The present work was able to show that there are differences among the PELD techniques.

Iprenburg et al state that the radiation doses required for TF-PELD are small and should not be enough to discourage patients from considering endoscopic spine surgery. They also state that L5-S1 PELD are significantly longer in duration than PELD at other lumbar levels and require a longer fluoroscopy period of exposure .[15] The present work did not consider L5-S1 PELD in a different group, since we believe that difficult and longer cases of TF L5S1 discectomies might be due to lack of anatomical and disease considerations that should favor the IL approach instead. There were 3 patients submitted to TF-PELD for L5S1 disc herniations, for whom the time and amount of radiation were among the whole group average (Pts 2, 12 and 18 on ►**Table 1**). On the other hand, Choi et al, in a multi-center study, reported no significant difference of the radiological features between the iliac crest and L5S1 disc space in two similar groups submitted either to TF or IL-PELD. Meaning that the surgical team used to perform the TF-PELD had similar patient characteristics when compared with the patients of the surgical team used to perform IL-PELD.[18,19] Nevertheless, high iliac crest, large transverse process, high upward migration and axillary type disc herniation are still challenging for L5S1 TF-PELD.[18] In regard to fluoroscopy, if TF-PELD is attempted in those cases, the amount of time and radiation exposure would certainly be higher. Along with other advantages, particularly in patients with wide interlaminar window, IL-PELD should be preferred.

Choosing the better approach for each patient, considering not only the surgeons preference[18] but the disease and anatomic particularities, may influence the amount of radiation exposure. While for some groups, L4L5 disc herniations may be considered for a TF-PELD approach only, the above-mentioned considerations should allow for a different approach.[20] Indeed, patient n14 of this group had an L4L5 posterolateral disc herniation and due to a wide L4L5 interlaminar window, whereas hypertrophic facets, was submitted to IL-PELD and needed an average amount of fluoroscopy (9 seconds and 13.86 mGy).

Conclusion

Interlaminar PELD requires a smaller amount and less time of radiation exposure than TF-PELD. Studies that concern radiation required for MISS should consider the PELD approaches separately.

Conflicts of Interest

The authors have no conflicts of interest to report.

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Double Crush Syndrome of the Median Nerve: A Literature Review

Síndrome do duplo impacto do nervo mediano: uma revisão de literatura

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Abstract

Double crush syndrome (DCS) is defined as the compressive involvement of the same peripheral nerve in different segments. When this syndrome affects the median nerve, a proximal compression of a spinal nerve that will constitute this structure (often the spinal nerve at the C6 vertebra) is usually noted at the cervical spine level as a herniated disc and as a distal compression at the level of the carpal tunnel. Epidemiological data on median nerve compromise by DCS are still very scarce in the medical literature. The diagnosis can be inferred by symptoms and signs occurring proximally and distally in the arm, as well as by alterations revealed by upper limb electromyography and neuroimaging studies, such as magnetic resonance imaging (MRI) of the cervical spine. Nowadays, information on which compressed neuroanatomical point should be initially addressed still depends on further studies. Limited data infer that these patients, when submitted to surgical treatment in only one of the median nerve compression points, evolve with worse functional outcomes than the surgically-treated group with carpal tunnel syndrome without DCS.

Keywords
- double crush syndrome
- median nerve
- carpal tunnel
- neuropathy

Resumo

A síndrome do duplo impacto (SDI) é conceituada como o comprometimento compressivo de um mesmo nervo periférico em segmentos distintos. Quando esta síndrome acomete o nervo mediano, usualmente se nota uma compressão proximal de um nervo espinhal que irá constituir o nervo mediano (frequentemente, o nervo espinhal da vértebra C6) ao nível da coluna cervical por uma hérnia discal e uma compressão distal ao nível do túnel do carpo. Dados epidemiológicos sobre a SDI comprometendo o nervo mediano ainda são muito escassos na literatura médica.

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Palavras-chave
- síndrome do duplo impacto
- nervo mediano
- túnel do carpo
- neuropatia

Observa-se que o diagnóstico pode ser inferido por sintomas e por sinais presentes proximal e distalmente no membro superior, além de alterações presentes na eletroneuromiografia de membros superiores, bem como achados em exames de neuroimagem como a ressonância magnética da coluna cervical. Atualmente, a informação sobre qual ponto neuroanatômico de compressão deveria ser abordado inicialmente ainda depende de mais estudos. Dados escassos inferem que esses pacientes, quando submetidos ao tratamento cirúrgico em apenas um dos pontos de compressão do nervo mediano, evoluem com resultados funcionais piores do que o grupo de pacientes com síndrome do túnel do carpo sem SDI tratados cirurgicamente.

Introduction

Double crush syndrome (DCS) refers to the coexistence of a double compression along the course of the same peripheral nerve.[1,2] A hypothesis proposed in 1973 by Upton et al describes the potential susceptibility of a distal nerve segment to lesion after a proximal injury.[1,3] It also emphasizes that the damage produced by double compression exceeds the additive injury caused by each compression, harming the global functioning of the nerve cells.[4,5]

This hypothesis can explain why patients with carpal tunnel syndrome (CTS) sometimes feel pain in the forearm, in the elbow, in the arm, in the shoulder, in the chest, and in the upper back region.[1,6] The DCS also can explain some cases of surgical failure, when the isolated CTS treatment did not result in pain control.[1,6] The DCS manifests as a disorder including pain, numbness and weakness due to two or more insults to the same peripheral nerve.[3,7] When it involves the median nerve, the coexistence of CTS and of cervical radiculopathy is the most frequent DCS pattern.[2,4]

CTS affects $\sim$ between 3 and 4% of the general population, with a prevalence of female patients in a 5:1 ratio, and in ages ranging from 30 to 60 years old.[8–10] The incidence of cervical radiculopathy appears to be in $<$ 1% of the population, often with a peak at the ages between 50 and 54 years old.[11–13]

In view of these issues, the present study aims to describe the epidemiological aspects, the risk factors, and the histopathological mechanisms involved in the DCS.

Methodology

The present study is a bibliographic review based on the specialized literature, and it was performed through the consultation of selected scientific papers at the PubMed database. To prepare the present work, the search used the descriptors *double crush syndrome* and *median nerve*. Only papers with patients presenting median nerve compression were included. Papers on distal compression of the median nerve with no CTS involvement were excluded.

Discussion

Carpal tunnel syndrome is the most frequent compressive syndrome, and it is defined by the compression of the median nerve at the level of the wrist. Its first description is assigned to Paget, who reported a case of medial nerve compression as a result of a distal radius fracture.[14]

The carpal tunnel is an inextensible osteofibrous tunnel defined as the space between the flexor retinaculum (FR), which constitutes its roof, and the carpal channel, which constitutes its fundus. It is delimited at the ulnar border by the hook of the hamate bone, the pyramidal bone and the pisiform bone, and at the radial border by the scaphoid bone, the trapezoid bone, and the FR. Its base is formed by the capsule, and the anterior radiocarpal ligaments cover the underlying portions of the scaphoid, semilunar, capitate, hamate, trapezium and trapezoid bones.[15] The median nerve is followed by four tendons of the digital flexor superficial muscles (FSD), four tendons of the digital deep flexor muscles (FPD), and by the tendon of the long flexor muscle of the thumb (FLP).[15] The FLP is the most radial element. At the entrance of the tunnel, the median nerve is dorsal to the tendon of the palmaris longus (PL) muscle, between the carpal radial flexor tendon (FCR) and of the digital superficial flexor tendon (SDFT).[15] At the distal part of the tunnel, the median nerve is divided in lateral, medial and thenar recurrent branches; the former two branches subdivide further.[15] The thenar branch passes through a separate tunnel before entering the thenar muscles in 56% of the cases.[15]

Typical CTS symptomatology is manifested by pain, hypoesthesia and paresthesia in the sensory innervation territory of the median nerve and, in some cases, by paresis for thumb opponency and abduction, as well as by signs of thenar hypotrophy.[2]

The differential diagnosis should include C6 and C7 vertebrae radiculopathy, proximal medial nerve compressions in the arm (Struthers ligament) or in the forearm (pronator syndrome), and double impact syndrome. Less frequently, upper motor neuron disease, cervical intraradicular injuries, neuropathies, and syringomyelia can be considered in the differential diagnosis.[2]

As already discussed, DCS is postulated as the compressive involvement of more than one point of the same peripheral nerve. In median nerve DCS, this can be exemplified by the compression of a spinal nerve—which will contribute to the formation of the median nerve at the brachial plexus level—and by the presence of an injury at a distal point of the same nerve, especially in the carpal tunnel.[1]

The prevalence of DCS in CTS patients, cumulated with cervical radiculopathy, varies greatly in the medical literature. Some studies report a 5% prevalence, while others state that 94% of the studied patients are affected.[16–18] This extensive variation could be partially explained by the lack of a gold standard for the diagnosis of DCS.[16–18] Some aspects, however, can be pointed out, such as the higher prevalence in males, according to studies by Tian et al.[19]

Different physiopathological mechanisms for DCS were suggested, and can be divided into highly plausible (1–4) and plausible.

(1) Axonal transport: Animal studies revealed that axonal transport is compromised by the mechanical pressure on the nerve at levels commonly observed in humans.[16]
(2) Super-regulation and under-regulation of ion channels: The super-regulation of sodium channels and the under-regulation of potassium channels are observed at points adjacent to the nerve compression site and could lead to a reduction in the neuronal action potential threshold.[16]
(3) Inflammatory process in the spinal nerve sensory ganglion: The mechanical compression process of the spinal nerve would lead to the invasion of its sensory ganglion by immune cells. This process would trigger the release of cytokines that would lower the threshold to initiate action potentials in sensory neurons located at the sensory ganglion.[16]
(4) Neuromas-in-continuity: Mechanical lesions on a peripheral nerve with an intact epineurium could lead to a defective regeneration of axon fibers in such a way that, during this process, they could not reach the peripheral target, forming neuromas along the nerve trunk.[16]
(5) Central sensitization: The nerve compression process would lead to central sensitization, promoting membrane excitability changes that would reduce inhibition, causing a reduction in the pain thresholds.[16]
(6) Nerve biomechanical aspects and movement patterns: During the movement of the limbs, the nerve slides over the adjacent tissue in longitudinal and transverse directions. Disturbances, such as a mechanical compression, could increase distal tension, compromising the biomechanics of the nerve.[16]
(7) Cognitive, psychological and psychosocial factors: Fear or hypervigilance states would reduce pain thresholds in these patients.[16]
(8) Peripheral nerve and spinal cord immunoinflammation: Immunoinflammatory processes at different points of the nervous system could reduce the action potentials thresholds of sensory neurons.[16]
(9) Microcirculation damage: The nerve compression would affect the microcirculation of the nerve, resulting in intraneural and extraneural edema.[16]
(10) Combined mechanisms: This theory tries to explain DCS through a sum of the aforementioned mechanisms.[16]

The risk factors for DCS include conditions that predispose patients to CTS and cervical disc herniation. The risk factors for CTS include diabetes mellitus, alcohol abuse, amyloidosis, rheumatoid arthritis, infectious synovitis, gout, dermatomyositis, scleroderma, hypothyroidism, long-term hemodialysis, obesity, repeated wrist flexion-extension movements, extended wrist during loaded movements, and fist injuries.[20] The risk factors for cervical disc herniation include smoking, cumulative effect of spine microtraumas and macrotraumas, and osteoporosis.[21] It is noteworthy that some studies point to the male gender and increased age as independent risk factors for DCS.[22]

The specific symptomatology observed in DCS patients are CTS symptoms plus pain and cervical region, shoulder and upper limb hypoesthesia ipsilateral to the nerve compression.[23]

The electromyographic changes observed in DCS are more pronounced than those present in CTS alone. A study comparing electroneuromyography (ENM) findings in patients with only CTS or DCS (CTS plus cervical radiculopathy) shows that the latter present lower sensory and motor nerve conduction velocity and higher distal motor latency. Importantly, these ENM differences were statistically significant.[23]

Although there are specific diagnostic criteria for CTS, they are yet to be established for DCS.[13,24] The diagnosis of cervical radiculopathy is determined by the clinical history of the patient and imaging tests, such as cervical X-rays, which can show cervical osteophytosis, as well as computed tomography (CT) and magnetic resonance imaging (MRI) exams.[25,26] For some authors, the history of the patients, signs such as Phalen or Tinel-Hoffmann, and the presence of thenar hypotrophy can be very relevant.[27,28] These clinical tests serve as screening tools for an electroneuromyographic study, which is considered the gold standard for the diagnosis of CTS.[23,28]

Regarding the most appropriate surgical treatment for DCS patients, some inferences can be suggested from studies with CTS patients with unsatisfactory postprocedural pain control.[4,29] The isolated surgical treatment of the distal compression did not provide pain control if there was a proximal compression of the median nerve. Eason described 47 CTS surgeries with unsatisfactory outcomes for pain control.[4,29] In 38 of these procedures, it was possible to identify an intervertebral foramen stenosis at the cervical level.[4,29]

Two remaining issues would be the best course for DCS patients, and which nerve compression point should be initially approached in surgery. Baba et al reviewed the medical records of 483 patients, from which 65 showed peripheral and cervical signs and symptoms, being classified as DCS carriers.[23,25] These authors suggest that the decompression of the cervical spinal nerve should be initially performed to reduce myelopathy risks due to cervical disc herniation.[23] As such, later, the patient would be submitted to CTS treatment.[23]

Conclusion

Although DCS is recognized as a distinct clinical entity, the exact operation of its neurophysiological and cellular mechanisms is not yet sufficiently known and universally accepted. Some patients with CTS can be DCS carriers. There is no consensus regarding the median nerve site that should be initially decompressed. Further studies are required in order to better document the physiopathology and the most efficient treatment for this condition.

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Learning Curve in Spine Surgery—Too Many Techniques, Too Many Options—How Should Young Surgeons Deal with this Anxiety?

Curva de aprendizado em cirurgias de coluna—Muitas técnicas, muitas opções—Como jovens cirurgiões devem lidar com a ansiedade?

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Spine surgeons have long years of training to get their licenses and start clinical practice. After a long medical school graduation and some years of neurosurgery or orthopedic residence, an additional fellowship is generally performed for 1, 2, or even 3 years. When the full license is obtained, young surgeons have to deal with another problem: the large amount of surgical options and techniques they have to learn. However, should we have to learn all surgical techniques?

Modern spine surgery is based on the understanding of the whole spinal alignment and also of the spinopelvic relationships, as well as on the understanding of the specific characteristics of the patients, such as bone quality, personal expectations, comorbidities, and others. Regarding degenerative lumbar spine surgeries, specifically, decompressive techniques are probably the most common procedures performed in the daily practice. For this, surgical options include open traditional laminectomies, lateral decompressions using paramedian approaches, pars decompression, tubular surgeries, endoscopic procedures (interlaminar or foraminal), among an infinitude of small variations described in the medical literature.[1–4] When we talk about spinal fusion, there is also a multitude of procedures available (posterior lumbar interbody fusion [PLIF], transforaminal lumbar interbody fusion [TLIF], anterior lumbar interbody fusion [ALIF], oblique lumbar interbody fusion [OLIF] and lateral lumbar interbody fusion [LLIF], among others), with specific advantages and disadvantages. An entire career is necessary in order to achieve expertise in these procedures.[5,6]

Nevertheless, in our humble opinion, there is no surgeon who is able to perform all of the spine surgery procedures with the same level of mastery. Experience requires performing the same procedure for a long time, and even for experienced and large-volume surgeons, mastering all of the techniques is humanly impossible. The industry and their representative surgeons put on tremendous amount of pressure for young surgeons to learn these new procedures, most of which are much more expensive than the old ones (especially because they are not yet popularized) and without a clear and well demonstrated scientific superiority.

So, how can one deal with all this scientific noise? With this unbelievable amount of medical literature? In fact, the anxiety exists indeed. Personally, we do not know the answer, but we would recommend that surgeons focus on surgical indications and on understanding the nuances of the patient involved in the treatment (expectations, clinical conditions, and body habits, among others). The magic sauce is understanding who needs surgery and for what reasons, and then master a technique (new or old) that will accomplish your goals with the most cost-effective strategy. If you have a good surgical indication, perform the technique that you feel more comfortable with and whichever you have more experience in. Throughout the years, you will be able to understand what you should incorporate or not in your

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practice. Always learning, but without anxiety. Then, slowly, you may differentiate clearly what is an economical market pressure in your practice from what is really better for your patients. Finally, do not feel guilty for not knowing everything. This happens to every surgeon, all the time.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Intradural Extramedullary Spinal Metastasis of a Kidney Cancer: Case Report

Metástase espinhal intradural extramedular de carcinoma renal: relato de caso

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Abstract

Objective To report a case of intradural extramedullary metastasis originated from a kidney neoplasm. Metastases in this topography occur in a low frequency, especially considering that the primary tumor was located along the urinary system.

Case Report A male adult begins to present with pain in the lower limbs and develops flaccid paraparesis of the pelvic limbs, also attacking the control of the sphincter. The patient had a previous history of right nephrectomy due to a kidney neoplasm. After investigation with imaging exams, a metastatic lesion was found to be the source of the symptoms. The patient was surgically treated by the neurosurgery team of the hospital.

Conclusion Cases like this are not common, and considering the low incidence of these cases and the nonspecific symptoms, such as pain, we do not always come up with the hypothesis of a metastasis in this topography. The surgical treatment, although it is a palliative feature, has an important part in maintaining the performance and the quality of life of the patient.

Keywords
- neoplasm metastasis
- kidney neoplasms
- intradural metastasis

Resumo

Objetivo Relatar um caso de metástase intradural extramedular de um carcinoma renal. Metástases nesta topografia são de baixa ocorrência, ainda mais tratando-se de um tumor primário no sistema urinário.

Relato de caso Paciente adulto do sexo masculino que inicia quadro de dor nos membros inferiores, evoluindo com paraparesia crural, tem piora do quadro, acometendo a função esfincteriana. O paciente possuía um histórico prévio de nefrectomia por neoplasia maligna em sítio renal. Após investigação com exames complementares,

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- metástase neoplásica
- neoplasias renais
- metástases intradurais

foi detectada a lesão metastática responsável pelos sintomas. A mesma foi tratada cirurgicamente pela equipe de neurocirurgia do serviço.

Conclusão Casos como este não são comuns e nem sempre nos levam a aventar a hipótese de metástase nesta topografia, frente à baixa incidência e ao início de sintomas muitas vezes inespecíficos, como dor. O tratamento, apesar de paliativo, tem um papel importante na qualidade de sobrevida do paciente.

Introduction

Cancer is currently a highly prevalent disease, and a metastatic disease occurs as the clinical picture progresses. Systemic neoplasms with distant metastasis to the spine range from 30 to 40%.[1,2] Considering the spine, intradural extramedullary metastases are rare and represent ~ 1% of the cases.[3–5] The age group with higher prevalence of cases is between 40 and 60 years old.[4] Metastatic dissemination is variable and related to the histological type of the primary tumor, as well as to its primary location.[4,5]

Schwannomas, meningiomas, and neurofibromas are the primary central nervous system (CNS) tumors that most frequently send metastatic implants to the intradural compartment,[3,6] while the major systemic neoplasms that metastasize to the spine are breast, lung, and prostate tumors.[4,7] As technology advances, such specific diagnoses are now possible and assist in the treatment.[8] Primary renal cancer is one of the most uncommon amongst this narrow group of neoplasms, thus the relevance of the following reported case.

Case Report

A 62-year-old man reporting bilateral lower limb weakness for some time. The condition worsened 10 days before, when he became incapable of walking and presented with bowel and bladder dysfunction. The initial physical examination confirmed an asymmetric paraparesis, with the left side being more affected (muscle strength graded 2 on left leg and 3 on the right), as well as knee jerk hyporeflexia, and preserved sensitivity.

Previous medical history: diabetes mellitus, chronic arterial hypertension. Right nephrectomy 4 years before, due to a malignant kidney neoplasm.

Through the anamnesis and a physical examination, medullary compression was the hypothesis inferred. In order to advance the investigation, a lumbar spine magnetic resonance imaging (MRI) exam was requested. (►**Figs. 1–5**)

The MRI showed an intradural extramedullary heterogeneous expansive lesion located posterior to the L3 vertebral body and glancing to the L3-L4 right foramen, probably corresponding to a schwannoma. In association, intradural hemorrhagic content is observed, extending from the lesion to the sacral region.

An L3-L4 laminectomy was performed, a middle line opening on dural topography, microscopically, a gray soften mass could be visualized. Using standardized microsurgical techniques, the lesion was completely removed without adverse events. The metastasis was confirmed by histological analysis.

During the postoperative recovery, the patient presented melena, therefore the gastroenterology team was requested to evaluate him as well. An upper endoscopy was performed, revealing an elevated duodenal lesion located in the middle third, just above the major duodenal papilla. The lesion measured ~15 mm, was rounded and depressible, and showed erosion on its surface. The patient was discharged from the hospital on the 9th day postsurgery, presenting

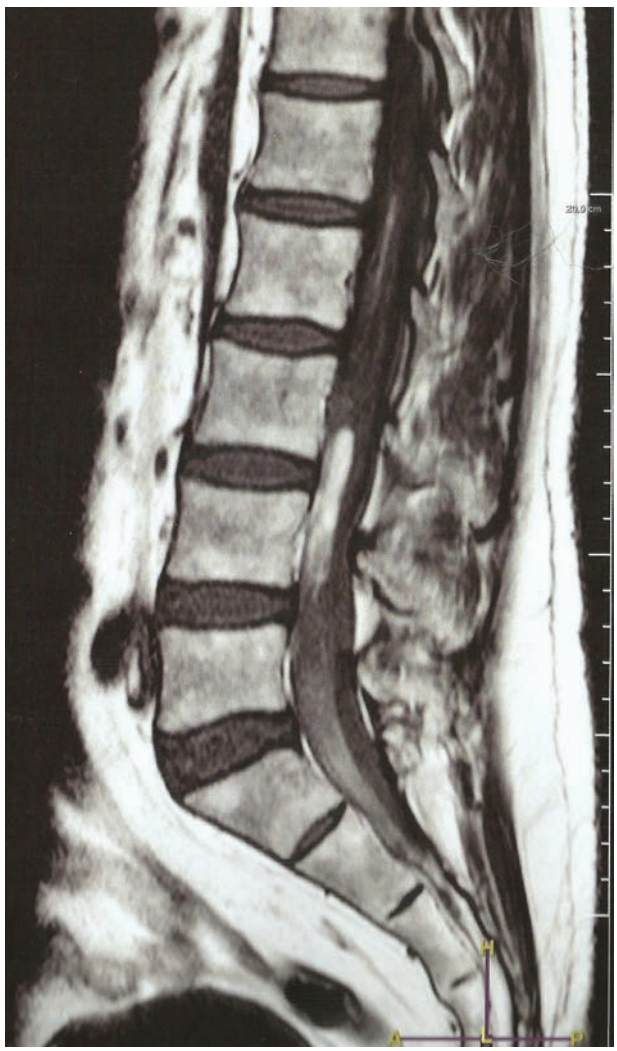

Fig. 1 Sagittal T1 lumbosacral magnetic resonance imaging showing an intradural extramedullary heterogeneous expansive lesion located posterior to the L3 vertebral body and glancing to the L3–L4 right foramen.

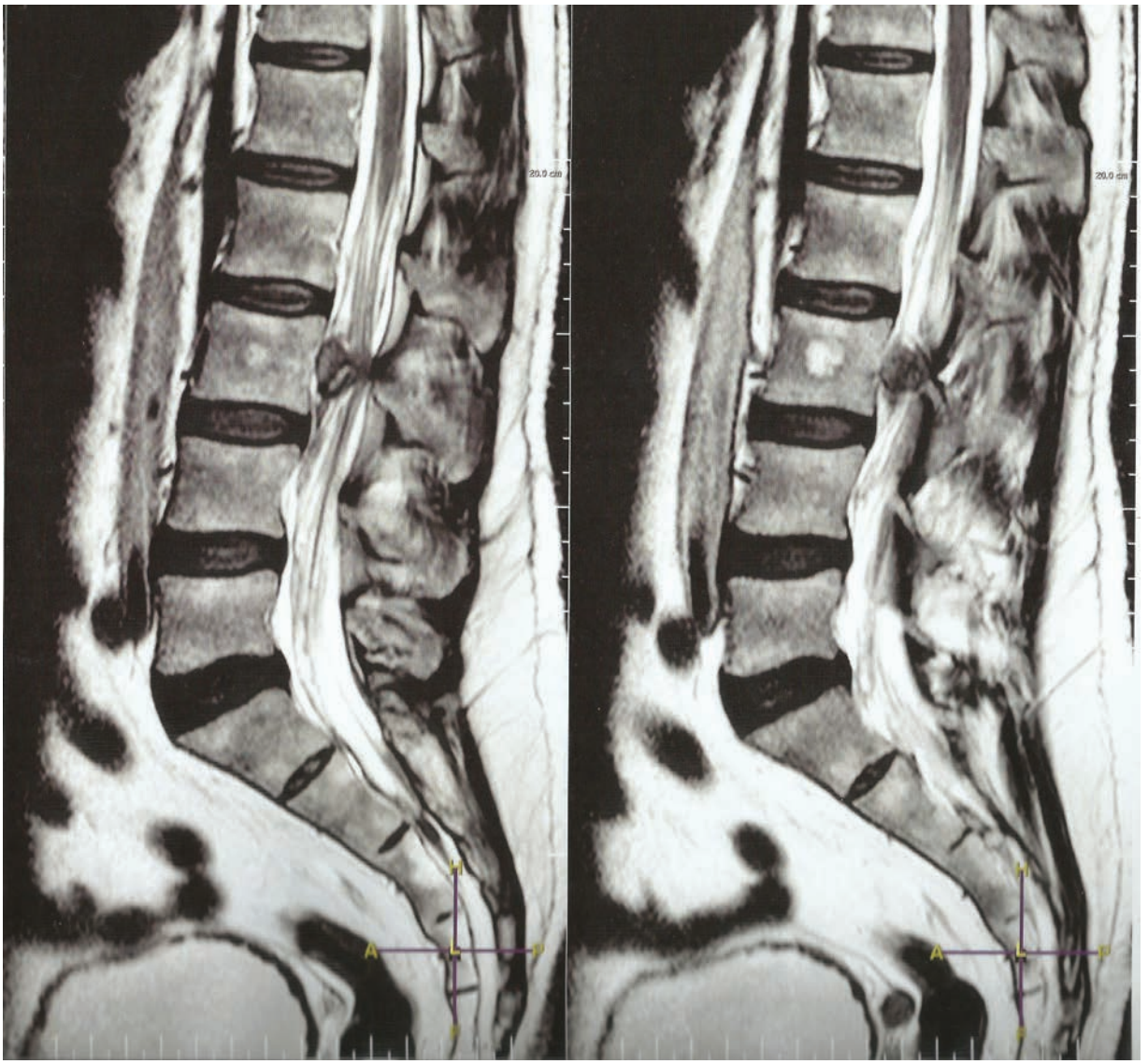

Fig. 2 Sagittal T2-weighted lumbosacral magnetic resonance imaging highlighting the intradural extramedullary heterogeneous expansive lesion located posterior to the L3 vertebral body and projecting to the L3–L4 right foramen.

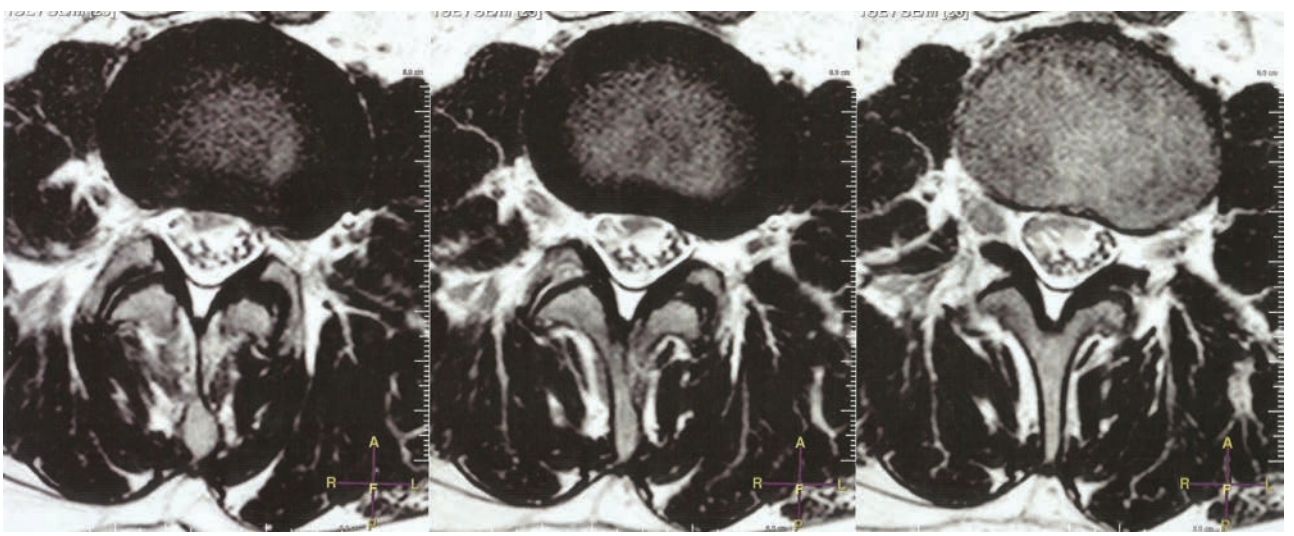

Fig. 3 Axial T2-weighted lumbosacral magnetic resonance imaging showing an intradural extramedullary expansive lesion glancing to the L3-L4 right foramen.

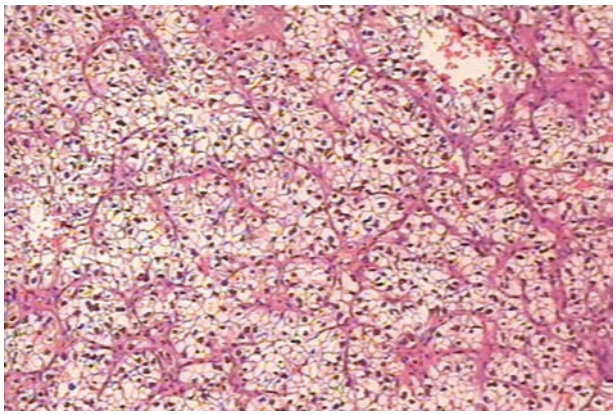

Fig. 4 Clear cell carcinoma. Neoplasic renal cells with highly lipidic and large cytoplasm encircled by small blood vessels. Hematoxylin and eosin stain, 100x magnification.

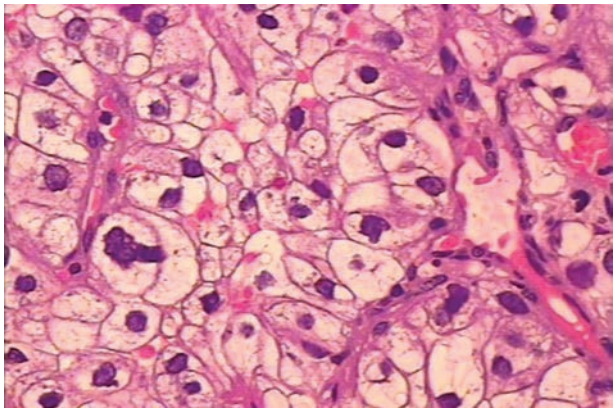

Fig. 5 Clear cell carcinoma. Neoplasic renal cells with highly lipidic cytoplasm, discreetly pleomorphic core. Hematoxylin and eosin stain, 400x magnification.

satisfactory strength recovery (muscle strength graded 3 on the legs and 4 on the thighs). Chemotherapy requirement was evaluated afterwards by the clinical oncology team.

Discussion

Metastatic dissemination to the intradural space includes: hematogenous, via the Batson plexus and the azygos vein,[4] by lymphatic extension, and even by contiguous invasion. When considering primary brain tumors and even synchronous brain metastasis, cerebrospinal fluid (CSF) dissemination must also be considered. This last type of metastatic dissemination may be related to the action of gravity; therefore, many extradural intramedullary metastases concentrate at the nerve roots of the cauda equina.[5,8] A study has proposed that perineural dissemination may also occur, spreading from the renal plexus through the sympathetic chain, and then to the intradural space, also affecting the cauda equina in some cases.[9]

The clinical features of spinal metastasis are vast and include symptoms due to local periosteal lesions, to adjacent tissue invasion, and, lastly, due to the compression of the roots of the nerves.[4] In general terms, the patients can present with pain, autonomic symptoms, motor and sensitive deficits. General signs may also be present, as in most consumptive systemic illnesses, such as weight loss, appetite loss, and sweating.[2,3] The intensity of the symptoms range in magnitude according to the aggressiveness of the tumor, which is represented by its replication rate, extension, and histological type.

The pain may be mechanical, radicular or local, and is the most common initial symptom, affecting up to 90% of the patients.[2,5] Several types of pain may be presented concurrently, since they have different mechanisms. Local pain is related to periosteal inflammation as a consequence of the tumoral growth; mechanical pain happens due to an osseous deformity that causes vertebral instability. As for the radicular type, it is related to the compression or irritation of the roots of the nerves.[2,7] In spite of their distinct sensitive features, or even of their absence, all of the pain types should lead to investigation.[2,7] Especially in patients with previous cancer diagnosis or even with just general risk factors to neoplasms, a precocious investigation leads to an early diagnosis and treatment.[10]

In the case reported in the present article, motor dysfunction was also present as part of the natural course observed in the typical clinical syndrome, originated by spinal metastasis. The progression of the syndrome may consist of cervicalgia and low back pain followed by sensory and motor impairment – with well delimited dermatomal distribution – and autonomic disturbance (erectile, bladder, and bowel dysfunction). The loss of muscle strength is critical and will advance to paralysis, unless proper early treatment is provided, especially considering intramedullary metastasis, which progresses even faster.[2] Autonomic disturbances are related to a worse prognosis and tend to appear only after 2 months after the onset of pain; this scenario highlights a delay in the diagnosis.[2]

Intradural extramedullary metastases tend to progress rapidly from pain to motor dysfunctions, and symptomatic patients are mostly classified in advanced stages of the disease.[11] A more precise investigation and staging is done to guide the treatment options. When assuming a case is likely metastatic, it is essential to intensify the investigation with additional methods to confirm the hypothesis and plan interventions more precisely. Amongst a vast amount of imaging methods currently available, MRI is considered the gold standard to assess spinal metastasis.[8] This method offers excellent results at a fair cost and is widely accessible, besides its superior sensitivity to evaluate soft tissues in detail, when compared with other imaging studies.[2,7]

The treatment for spinal metastasis is palliative, as its main goal is to relief discomfort and pain, improving the quality of life of the patient.[4,5] The estimated survival time postdiagnosis is of between 4 and 7 months.[11] The treatment options vary from radiotherapy to chemotherapy, surgical decompression, and even hormonal therapy. The therapeutic modality is chosen based on the performance status and clinical stability of the patient, and on if the life expectation is inferred to be $>$ 3 months.[2,6,7] Some authors have suggested that, regarding the possible association between metastasis and primary CNS tumors, it may be appropriate to perform neuraxial chemotherapy and radiotherapy concomitantly.[5]

Surgical therapy is an acceptable choice that improves the quality of life, but has no effect in prolonging survival[11]

In our case, surgical therapy was the best option, considering the previous good performance status of the patient, the fast advancing muscle weakness, and also the availability of an experienced neurosurgery team in a well-equipped hospital. Surgery can also be considered as a valid treatment modality, showing benefits especially when intractable pain is part of the case, or to improve intense neurological symptoms, therefore improving the quality of life.[11,12] In clinical cases involving the cauda equina or tumors without cleavage plan, we should not aim for total gross resection, since a decompressive laminectomy may offer a satisfactory result with lesser risks.[13] A new possibility in surgical modality is stereotactic radiosurgery, which shall target the tumoral cells and preserve healthy tissues, benefiting patients with high surgical risk and also those who remain with residual disease postsurgery.[13] Still, little research has been done to elucidate the outcomes in patients submitted to this treatment.[13]

Conclusion

Cases involving intradural extramedullary spinal metastases are not commonly seen in the clinical practice, particularly when the primary site of the tumor is the kidney. Metastases themselves are indicatives of advanced disease, but considering the spinal impairment, the treatment can be palliative only. Surgery aims to reduce pain symptoms and to improve the quality of life of the patient. Comparative studies regarding specific indications to the modality of the surgery, as well as technology advances in oncologic therapies, are necessary so that the treatment improves and we move forward toward better results for our patients.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Superior Sagittal Sinus Invasion by Malignant Glioma: Case Report and Literature Review

Invasão do seio sagital superior por glioma maligno: Relato de caso e revisão da literatura

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Abstract

Anaplastic oligodendrogliomas (AOs) correspond to ~ 23% of all oligodendrogliomas. They correspond to a tumor with malignant histological characteristics, focal or diffuse, associated with a worse prognosis. In the present case report, we describe the case of a 30-year-old female submitted to resection of a right parietal lesion whose histology showed to be an AO. She underwent complementary treatment with chemotherapy and radiotherapy according to the Roger Stupp protocol. Four years after the initial diagnosis, there was tumor recurrence within the superior sagittal sinus, with no evidence of recurrence elsewhere. In the literature, we have found no similar published case reinforcing the rarity of this condition

Keywords
- anaplastic oligodendroglioma
- superior sagittal sinus

Resumo

Oligodendrogliomas anaplásicos correspondem a ~ 23% de todos os oligodendrogliomas. Eles correspondem a um tumor com características histológicas malignas, focais ou difusas, associadas a um pior prognóstico. Descrevemos aqui o caso de uma mulher de 30 anos de idade submetida à ressecção de uma lesão parietal direita, cuja histologia mostrou ser um oligodendroglioma anaplásico. A paciente foi submetida a tratamento complementar com quimioterapia e radioterapia de acordo com o protocolo de Roger Stupp. Quatro anos após o diagnóstico inicial, ocorreu recidiva tumoral no seio sagital superior, sem evidência de recorrência em outro local. Na literatura, não encontramos nenhum caso semelhante publicado reforçando a raridade desta condição.

Palavras-chave
- oligodendroglioma anaplásico
- seio sagital superior

Introduction

Oligodendrogliomas represent the 3rd most common type of glial tumors, corresponding to between ~ 4 and 15% of all gliomas of the central nervous system (CNS).[1]

Anaplastic oligodendrogliomas (AOs) (World Health Organization [WHO] grade III)[2] correspond to ~ 23% of all oligodendrogliomas.[3] They correspond to a tumor with malignant histological characteristics, focal or diffuse, associated with a worse prognosis,[1] in comparison to the oligodendrogliomas WHO grade II.[2] Anaplastic oligodendrogliomas are associated with a 5- and 10-year average survival rate of 63% and 33%, respectively.[4]

In the present case report, we describe the case of an AO with a pattern of tumor dissemination not described in the literature, into the superior sagittal sinus (SSS), with its consequent obliteration, in a patient with no evidence of tumor recurrence elsewhere.

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Case Report

A 30-year-old woman presented with an inaugural seizure in 2013. She was submitted to a computed tomography (CT) of the brain that revealed a right parietal corticosubcortical lesion, with calcifications (►**Fig. 1**), and to a magnetic resonance imaging (MRI) of the brain that revealed a hypointense lesion in the T1 weights with areas of contrast enhancement (►**Fig. 2**) suggesting a high-grade glioma. The patient underwent a surgical excision with gross total resection (GTR) of the lesion. In the postoperative period, she developed a left hemisphere apraxia and a left inferior quadrantanopsia.

The neuropathological study showed a highly cellular glial tumor, with diffuse invasion of the cerebral parenchyma, composed of cells of relatively uniform round nuclei and with frequent perinuclear halos (►**Fig. 3**). Necrosis, frequent mitoses, and a moderate proliferation index were also observed. The immunohistochemistry study was positive for glial fibrillary acidic protein (GFAP), and the tumor was isocitrate dehydrogenase (IDH-1) and α thalassemia/mental retardation syndrome X-linked (ATRX) positive. Based on these characteristics, the diagnosis was AO (World Health Organization grade III[2]).

The complementary treatment was started according to the Roger Stupp protocol. The patient completed 30 sessions of radiotherapy (2Gy/session, in a total of 60 Gy), with concomitant temozolomide 75mg/m^2 for 7 days per week, for 6 weeks. After that, she underwent 6 cycles of adjuvant temozolomide 150mg/m^2 for 5 days per month.

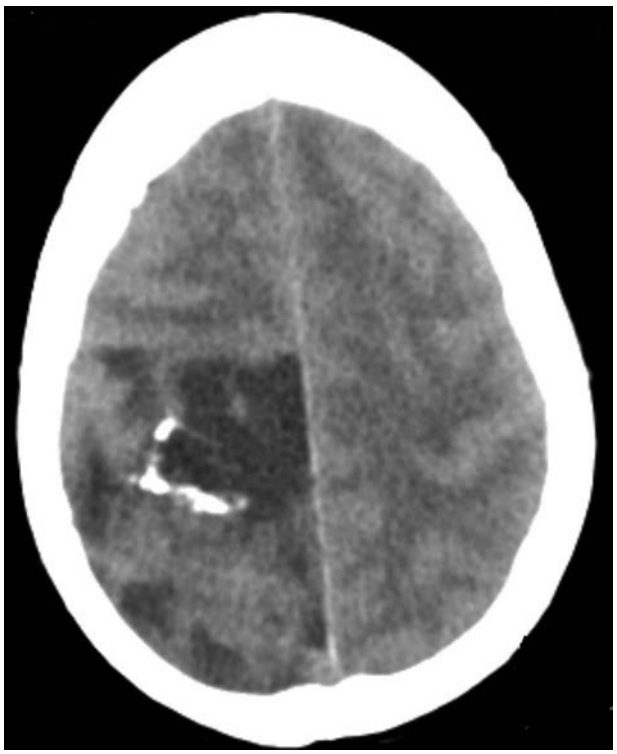

Fig. 1 Computed tomography scan showing a right parietal lesion with calcification.

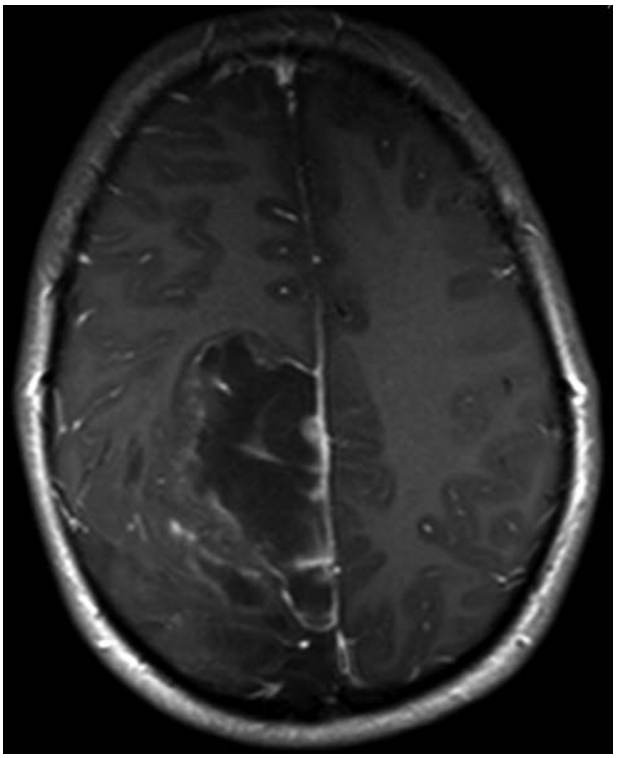

Fig. 2 Axial T1-weighted magnetic resonance imaging showing a hypointense right parietal lesion with areas of contrast enhancement.

Four years after the initial diagnosis in an imaging control by MRI, a recurrence of the tumor was observed, located within the SSS in the middle and posterior third, with no recurrence elsewhere, particularly in the surgical site (►**Fig. 4**). Considering the hypothesis of surgical reintervention, an angiography was performed, which confirmed the absence of blood flow in the invaded portion of the SSS (►**Fig. 5**).

The patient was submitted to a new surgical intervention with en bloc resection of the occluded SSS, and it was verified intraoperatively that the external walls of the sinus were not invaded by the tumor and had a cortical vein that drained to

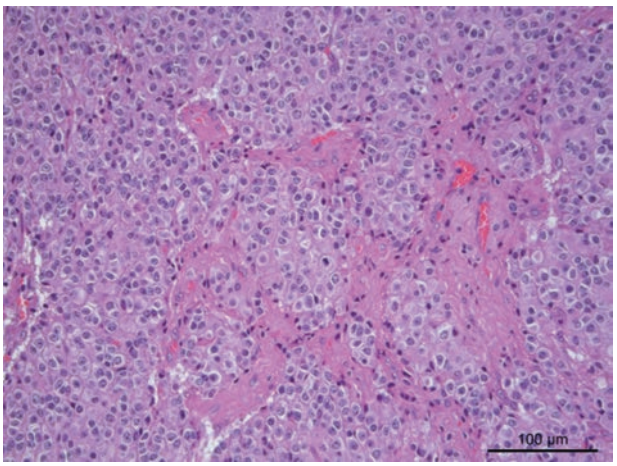

Fig. 3 Hematoxylin and eosin staining showing a hypercellular tumor with round nuclei with a perinuclear halo, with frequent mitoses.

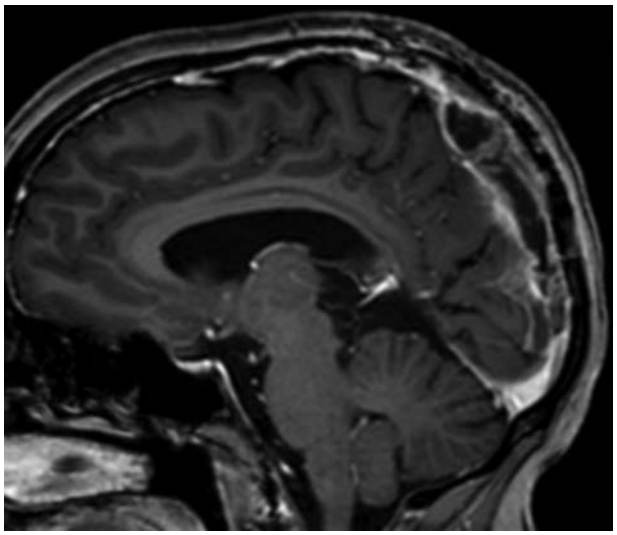

Fig. 4 Sagittal T1-weighted magnetic resonance imaging with contrast showing tumor recurrence located within the superior sagittal sinus in the middle and posterior third.

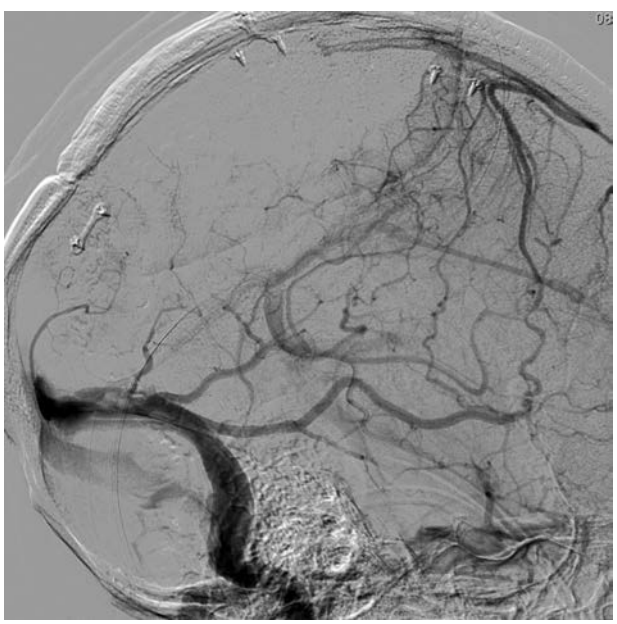

Fig. 5 Angiography with absence of flow in the middle and posterior third of the superior sagittal sinus.

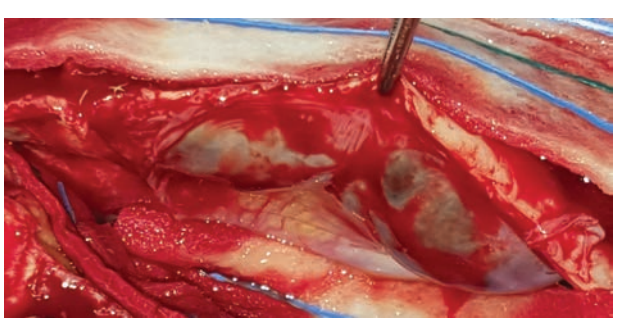

Fig. 6 Intraoperative images showing the sinus walls macroscopically without tumor invasion and a cortical drainage vein in the direction of the occluded superior sagittal sinus.

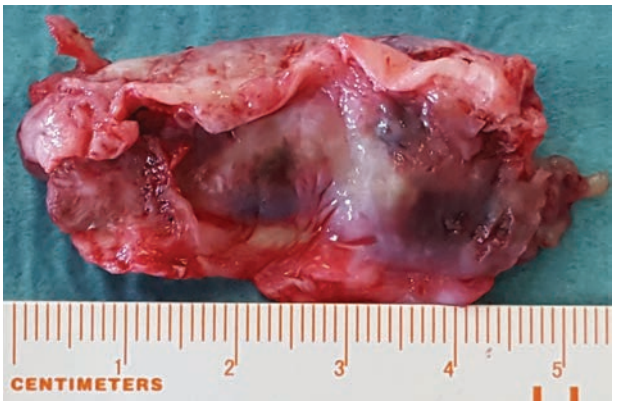

Fig. 7 En bloc resection of the tumor.

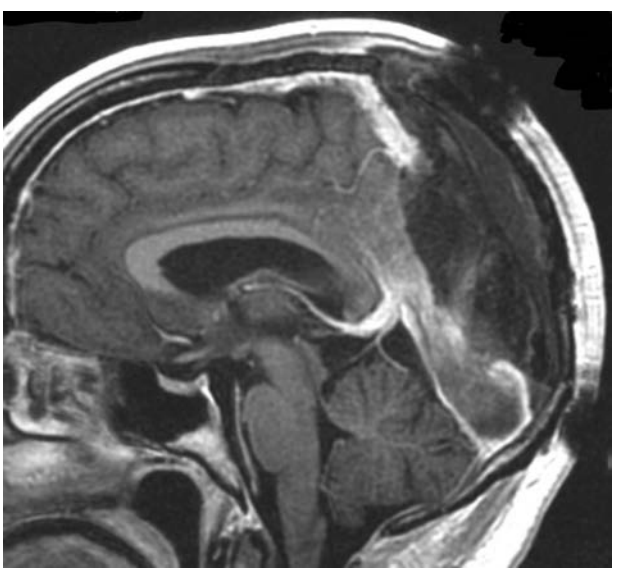

Fig. 8 Second surgery postoperative sagittal T1-weighted magnetic resonance imaging showing the resection of the occluded sinus.

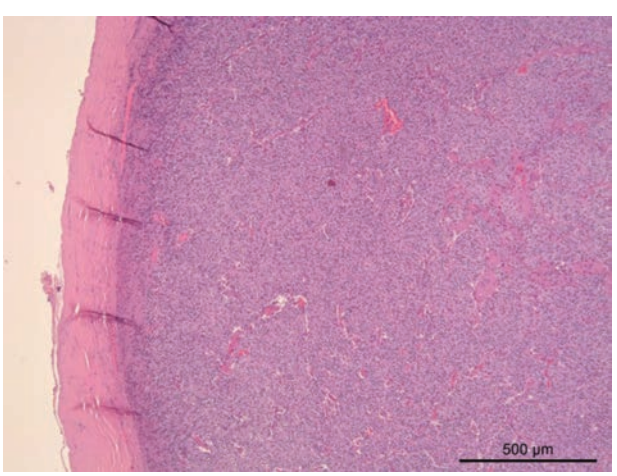

Fig. 9 Hematoxylin and eosin staining showing a hypercellular tumor, but no invasion of the sinus wall by malignant cells.

the occluded portion of the SSS (►**Figs. 6** and **7**). A postoperative MRI was performed, which showed the total removal of the invaded portion of the sinus (►**Fig. 8**). The neuropathological study showed a tumor with the same characteristics as the previous tumor, and the walls of the sinus were not invaded by the tumor (►**Fig. 9**).

Currently, the patient presents 5 years of global survival, with status 1 in the Eastern Cooperative Oncology Group (ECOG) Scale of Performance Status.

Discussion

Oncological diseases are often associated with hypercoagulable states due to the prothrombotic properties of neoplastic cells.[1] Malignant tumors of the CNS, mainly gliomas, are associated with deep venous thrombosis and pulmonary thromboembolism.[1,5] Several cases of venous sinus thrombosis associated with malignant CNS tumors and complementary treatments have been described in the literature.[6]

The case described in the present report refers to a tumor that spread to the interior of a cerebral venous sinus, without continuity spread, since the outer wall of the sinus was not invaded and there was no evidence of disease progression elsewhere, particularly in the surgical site. From the research done in the PubMed and Google Scholar databases, we have found no registered case of exclusive dissemination to the venous sinuses of a glial tumor.

We believe that because glial tumors are an oncology pathology, a state of hypercoagulability may have occurred, which in turn has caused a decrease in the SSS drainage flow. This fact created favorable conditions for the tumor cells to spread to the interior of the sinus through cortical veins and to find conditions to fix in and metastasize to the SSS.

Conclusion

Although no case is described in the literature, glial tumors can spread into the venous sinuses without further progression of the disease, as we have described in the present case.

In the case of absence of blood flow of the venous sinuses, in the context of CNS oncological pathology, we must always take into account in the differential diagnosis the possibility of obstruction caused by tumor invasion, especially in glial tumors.

Conflicts of Interests

The authors have no conflicts of interests to declare.

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Aneurysmal Bone Cyst of the Skull Base—Case Report

Cisto ósseo aneurismático da base do crânio—relato de caso

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Abstract

Introduction Aneurysmal bone cysts (ABCs) are pseudotumoral bone lesions of unknown etiology that are also hypervascularized, benign, and locally destructive. They are rare in the base of the skull. The present case report describes a case of aneurysmal bone cyst in the sella turcica.

Case Report The present study was developed at the department of neurosurgery of the Hospital Universitário Professor Alberto Antunes of the Universidade Federal de Alagoas (HUPAA-AL, in the Portuguese acronym), Maceió, state of Alagoas, Brazil, and is accompanied by a review of the literature from the PubMed database. A 17-year-old female patient with bitemporal hemianopia and intense left hemicranial headache associated with symptoms from the cranial nerves contained in the cavernous sinus. Neuroimaging evidenced a large lesion in the suprasellar region with calcification foci, sellar erosion, and extension to the cavernous sinus. The patient was submitted to a partial lesion resection and the histopathological analysis showed an aneurysmal bone cyst.

Conclusion A rare case of intracranial aneurysmal bone cyst, with the important differential diagnosis from pituitary adenoma.

Keywords

- aneurysmal bone cysts
- sella turcica

Resumo

Introdução Cistos ósseos aneurismáticos (COAs) são lesões ósseas pseudotumorais, de etiologia desconhecida, hipervascularizadas, benignas, localmente destrutivas. Cistos ósseos aneuristmáticos são raros na topografia da base do crânio. O objetivo do presente relato de caso é descrever um caso de cisto ósseo aneurismático localizado na sela túrcica.

Relato de caso O caso foi acompanhado no Serviço de Neurocirurgia do Hospital Universitário Professor Alberto Antunes da Universidade Federal de Alagoas (HUPAA-AL), Maceió, AL, Brasil, e sua descrição foi feita conforme dados encontrados em revisão de literatura realizada por meio do banco de dados PubMed. Paciente do sexo feminino, 17 anos, com hemianopsia bitemporal e intensa cefaleia hemicraniana esquerda

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associada à lesão dos nervos cranianos contidos no seio cavernoso. Exames de neuroimagem evidenciaram uma lesão extensa na região suprasselar com focos de calcificação e erosão da sela túrcica, e com extensão para o seio cavernoso. Tratamento com ressecção parcial da lesão, a qual o exame histopatológico revelou ser cisto ósseo aneurismático.

Conclusão Caso raro de cisto ósseo aneurismático intracraniano, sendo importante o diagnóstico diferencial com adenoma hipofisário.

Palavras-chave
- cistos ósseos aneurismáticos
- sela túrcica

Introduction

Aneurysmal bone cysts (ABCs) are pseudotumoral bone lesions of unknown etiology that are also hypervascularized, benign, and locally destructive; in addition, they often grow progressively. The literature reports an incidence of 0.14 per 100,000 individuals, corresponding to ∼ 1% of all bone tumors.[1] The long bones and the spine are the most commonly affected sites. Aneurysmal bone cysts are rare and more prevalent in children and in young people; they can evolve asymptomatically, or rapidly cause pain and neurological symptoms. The present case report describes a case of ABCs unusually located in the sella turcica.

Material and Methods

The present case report was described by the authors from the Neurosurgery Service of the Hospital Universitário Professor Alberto Antunes of the Universidade Federal de Alagoas (HUPAA-AL), Maceió, state of Alagoas, Brazil. The informed consent form was signed by a legal representative of the patient. The literature review was performed at the PubMed database up to December 2015.

Case Report

The patient J. F. S., female, 17 years old, was admitted to the HUPAA-AL with a clinical history (∼ 7 months) of mild to moderate hemicranial headache associated with visual field deficits. The condition worsened after 4 months, with severe left hemicranial headache. Upon admission, at the neurological examination, the patient was awake, a little confused, with no limb motor and/or sensory deficit, presenting left-side ptosis, anisocoric pupils (left > right), direct and consensual photomotor reflex absent on the left side and present on the right side, diplopia, fourth and sixth cranial nerve palsy on the left and right side, respectively, and bitemporal hemianopsia at the confrontation test, confirmed with a visual field test. After admission, the patient progressed with a stable clinical/neurological condition until she presented with an episode of psychomotor agitation associated with psychotic symptoms and delirium. She was submitted to a computed tomography (CT) of the head, which showed a sellar lesion with expansion and bilateral involvement of the cavernous sinus (►**Fig. 1**) and erosion of the upper portion of the clivus (►**Fig. 2**) and of the sellar floor (►**Fig. 3**). A magnetic resonance imaging (MRI) of the skull was not available during the hospitalization of the patient.

The results of routine laboratory tests, as well as of hormonal tests requested due to a suspected diagnosis of pituitary adenoma, were within the normal range.

A transsphenoidal approach aided by a surgical microscope was selected. The sellar and suprasellar lesion was partially resected. Maximal resection was not possible due to the location of the lesion and to the occurrence of major bleeding during the procedure.

In the postoperative period, the patient maintained the same clinical picture, with no signs of cerebrospinal fluid (CSF) fistula and/or incisional bleeding. The results of the anatomopathological study revealed a fibrovascular tissue composed of fusiform and epithelioid cells (giant cells) with fibrous tissue struts and hematic content consistent with an aneurysmal bone cyst (►**Figs. 4** and **5**).

The patient was lost at follow-up and the treatment outcome is unknown.

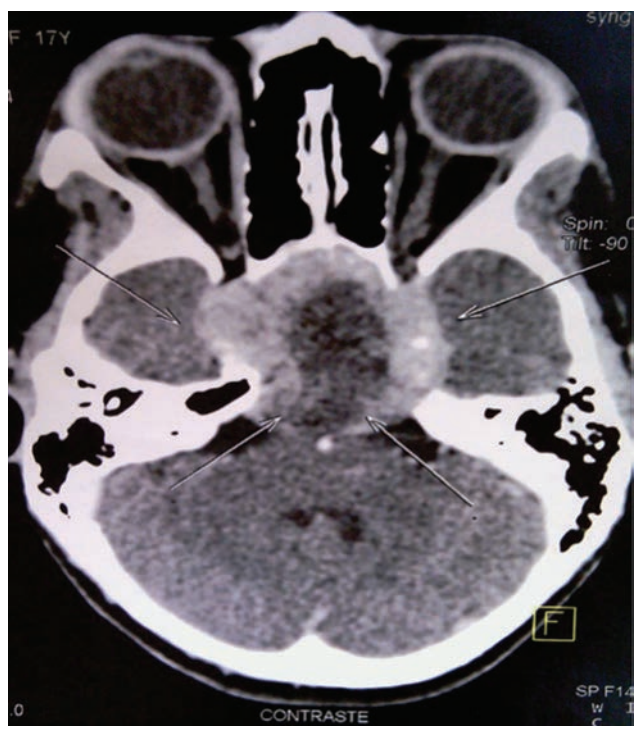

Fig. 1 Contrast-enhanced, axial head computed tomography. Expansive process at the base of the skull with sellar and parasellar regions involvement, cavernous sinuses invasion and apical clival and clinoid erosion; this lesion presents mixed density, predominantly with peripheral hyperdensity and central hypodensity.

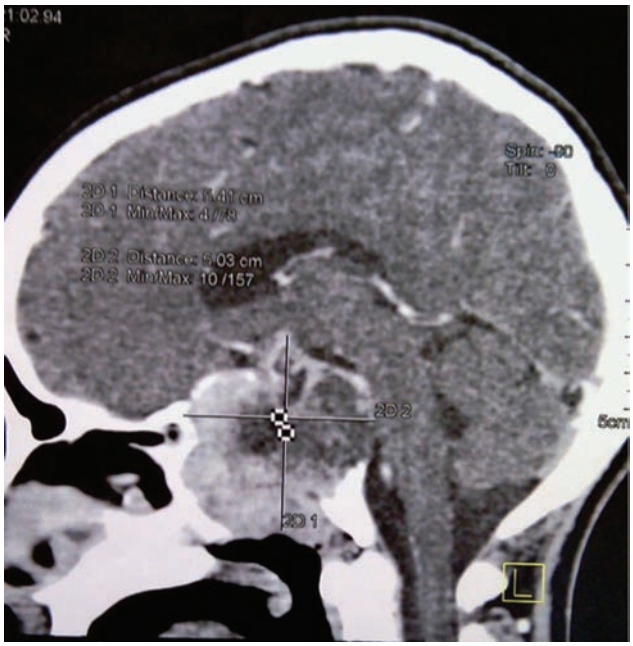

Fig. 2 Contrast-enhanced, sagittal head computed tomography. Expansive process of massive volume eroding the sella turcica and the base of the skull with inferior extension to the sphenoidal sinus and progressing to the clivus and cavum region.

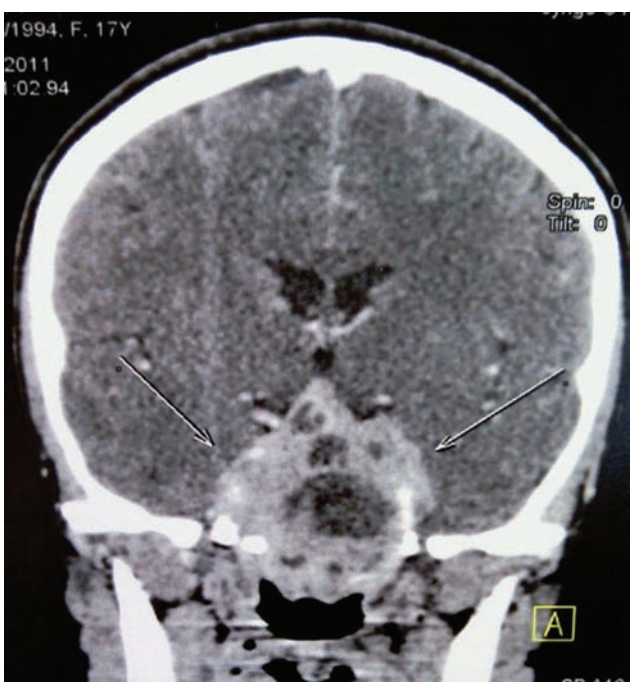

Fig. 3 Contrast-enhanced, coronal head computed tomography showing a tumor mass with invasive aspect, bilateral involvement of the cavernous sinuses and superior extension to the optic-chiasmatic cistern.

Literature Review

First described by Jaffe et al in 1942, ABCs are pseudotumoral bone lesions of unknown etiology that are hypervascularized, benign, locally destructive and grow progressively.[2] According to Lipman et al and to Biesecker et al, ABCs are non-neoplastic lesions composed of cystic cavities with

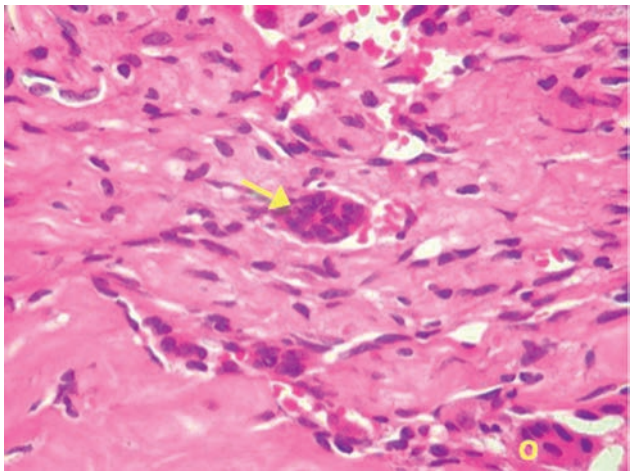

Fig. 4 This histological section (magnification 100x) shows an epithelioid cell (arrow), osteoclasts (O) and fusiform cells. Hematoxylin and eosin stain.

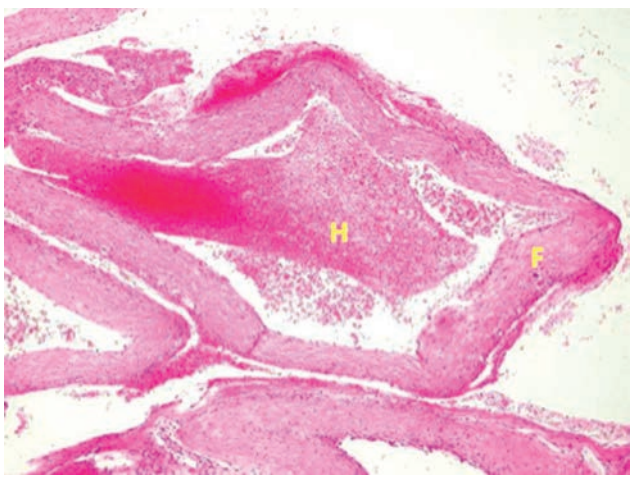

Fig. 5 This photograph shows the hematic content (H) within the fibrous septa (F). Magnification 40x. Hematoxylin and eosin stain.

fibrous walls and free blood circulation without endothelial delimitation.[3,4] Some older studies suggested that the origin of the lesion was the hemorrhagic degeneration of preexisting bone lesions (including giant cell tumor, osteoblastoma, fibrous dysplasia and chondromyxoid fibroma).[2,5,6] Isolated lesions are called primary lesions, whereas lesions arising from previous lesions are classified as secondary lesions.[7] In the present case, the nature of the lesion, whether primary or secondary, could not be established.

Lesions are macroscopically described as lytic bone structures with a thinner, brownish and fragile cortex (hematopoietic content),[8] and fibrous walls resembling soap bubbles.[9] Under light microscopy and routine staining (hematoxylin and eosin), cystic structures with fibrous or osseous septations containing fragile, hemorrhagic vascular tissue are described.[10] Some authors reported that the cyst is surrounded by hemosiderin, fibroblasts, giant cells, and stroma cells, but not by endothelium;[3,11] however, O'Brien et al, in 1994, reported the presence of endothelium and giant cells.[12] Since routine staining is sufficient for the diagnosis, immunohistochemical analysis is not required.[7,12]

Aneurysmal bone cysts are more commonly found in young individuals, and ~ 80% of the cases are diagnosed during the first 2 decades of life,[13] with a slight prevalence in female patients.[14,15] The most common sites for ABCs are the tibia, the femur, the humerus, the spine, and the pelvis.[5,6] Few studies show intracranial lesions, observed in only between 2 and 6% of all of the cases; the calvaria is the most common affected site in the skull.[16,17] The bones most commonly affected in the skull are the temporal (22%), the occipital (20%), the frontal (14%), the parietal (11%), the orbit (11%), the sinus (5%), the sphenoidal sinus, and the ethmoid bones (3%).[18–20] Only 15 cases located in the ethmoid bone were described up to 2014,[21–32] and 15 cases located in the sphenoid bone[26,30,33] were described up to 2015.

In some cases, the diagnosis of aneurysmal bone cyst is difficult because the radiological lesions are similar to other benign or malignant conditions, especially in unusual sites such as the base of the skull.[20,34]

In the present case, an important differential diagnosis to be considered at the neurological examination would be hypothalamic-pituitary-axis lesions. However, the radiological aspect of the lesion was a massive cystic formation with a solid portion spontaneously denser than the cerebral parenchyma and sellar bone erosion,[35] not corresponding to the description of the neuroimaging aspect of most pituitary macroadenomas. These lesions are homogeneous, and their density is similar to the parenchyma, except in cases with apoplexy and posthemorrhagic cystic cavities formation, which become similar to the one reported here.

Another differential diagnosis regarding imaging and topography would be craniopharyngioma, whose formation is similar to a cystic cavity; however, the aneurysmal bone cyst has thicker and irregular walls, formed by fibrous septations,[10] while the craniopharyngioma is a thin-walled cystic formation with calcification at the sellar base.

Treatment is performed by lesion curettage, followed by the injection of bone substitutes such as polymethylmethacrylate. Until the 1990s, the procedure included only lesion curettage; however, it was found that grafting reduced the recurrence rate from 26 to 17%, as mentioned by Mankin et al in 2005. These authors reported a recurrence rate in their own series of ~ 22%,[36] but other investigators found values ranging from 5 to 40%,[37] regardless of the approach.

Currently, the treatment of choice, whenever possible, is the complete lesion resection preceded or not by embolization.[7] Unfortunately, in the present case, this was impossible due to the location at the base of the skull and to the occurrence of a massive intraoperative bleeding. Some authors recommend radiotherapy, although its recurrence rate is > 30%,[7] while others contraindicate it due to the risk of sarcomatous degeneration.[24] There are also reports of interferon α-2a use in lesions considered unresectable.[29]

Conclusion

The present case report brings to light the knowledge of aneurysmal bone cysts in unusual topographies, such as the base of the skull, requiring its differential diagnosis with typically hypothalamic-pituitary-axis lesions, such as pituitary adenoma and craniopharyngioma. Although the treatment of choice is complete resection, it is not feasible at this location.

Conflicts of Interest

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Kernohan-Woltman Notch Phenomenon—Case Report

Fenômeno do entalhe de Kernohan-Woltman—relato de caso

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Abstract

The Kernohan-Woltman notch phenomenon is a paradoxical neurological manifestation consisting of a motor deficit ipsilateral to a primary brain injury. It has been observed in patients with brain tumors and with supratentorial hematomas. It is considered a false localizing neurological sign. Magnetic resonance imaging (MRI) scan has been the test of choice. The recognition of this phenomenon is important to prevent a surgical procedure on the opposite side of the lesion. The present case report describes a case of chronic subdural hematoma with a probable finding of the Kernohan-Woltman phenomenon, and it discusses its pathophysiology, imaging findings, treatment, and prognosis.

Keywords
- cerebral peduncle
- head injury
- Kernohan-Woltman notch phenomenon
- transtentorial herniation

Resumo

O fenômeno do entalhe de Kernohan-Woltman é uma manifestação neurológica paradoxal que consiste em um déficit motor homolateral à lesão cerebral primária. Este fenômeno tem sido observado em casos de neoplasia cerebral e de hematomas supratentoriais, e é considerado um sinal de falsa localização neurológica. Ressonância magnética tem sido o exame de escolha. É importante o reconhecimento deste fenômeno para evitar um procedimento cirúrgico no lado oposto à lesão. O presente relato de caso apresenta um caso de hematoma subdural crônico, com provável achado do fenômeno de Kernohan-Woltman, e discute sua fisiopatologia, seu achado de imagem, seu tratamento e prognóstico.

Palavras-chave
- pedúnculo cerebral
- traumatismo cranioencefálico
- fenômeno do entalhe de Kernohan-Woltman
- herniação transtentorial

Introduction

The Kernohan-Woltman phenomenon or Kernohan-Woltman notch signal is a hemiparesis ipsilateral to a supratentorial brain injury[1–5] in a brain tumor patient. The ipsilateral hemiparesis is secondary to a transtentorial herniation due to a midline deviation at the level of the midbrain, resulting in the compression of the contralateral cerebellar tentorium pyramidal fibers.[2] This leads to partial or total damage of these pyramidal fibers as they course the spinal cord to innervate the other side of the body. Injuries in these fibers cause a hemiparesis ipsilateral to the brain lesion, constituting a false localizing neurological sign.[2,5,7]

The present case report describes a probable case of Kernohan-Woltman phenomenon diagnosed through neurological examination and computed tomography (CT) scanning in a patient with a chronic subdural hematoma, as well as the physiopathology, diagnosis and prognosis of the condition.

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Case Report

A male patient, J. J. B., 59 years old, rural worker, presented with a history of mild head trauma 40 days before. The patient evolved with a moderate headache that was relieved with simple painkillers. For 72 hours, the patient had been presenting with a decrease in the level of consciousness, as well as decreased movements on the left side of the body and generalized seizures. The physical examination determined that the patient was in a regular general condition. The patient had systemic arterial hypertension (SAH), with no proper diet, but with sporadic use of specific medication. His blood pressure was 170 × 110 mm Hg. The neurological examination detected a decreased level of consciousness. At admission, the electrocardiogram (EKG) score was 10. The patient presented with left hemiparesis with crural predominance. The left pupil could not be viewed (►**Fig. 1**), and the right pupil measured 3.0 mm and presented little reaction to light stimuli. A CT of the head with no contrast medium revealed the absence of the left eye, as well as the presence of a double-density chronic subdural hematoma located in the left frontoparietal region with mass effect, compressing the ipsilateral brainstem, obliterating the homolateral ventricle and deviating midline structures (►**Figs. 2** and **3**). The patient was submitted to a left posterior parietal trepanopuncture and to drainage of the hematoma. Forty-eight hours after the surgical procedure, the patient was awake, with EKG = 13 and improvement of the left-sided muscle strength deficit. The seizures were controlled with hydantoin. The patient was discharged at the 6th postoperative day, with orientation for outpatient return.

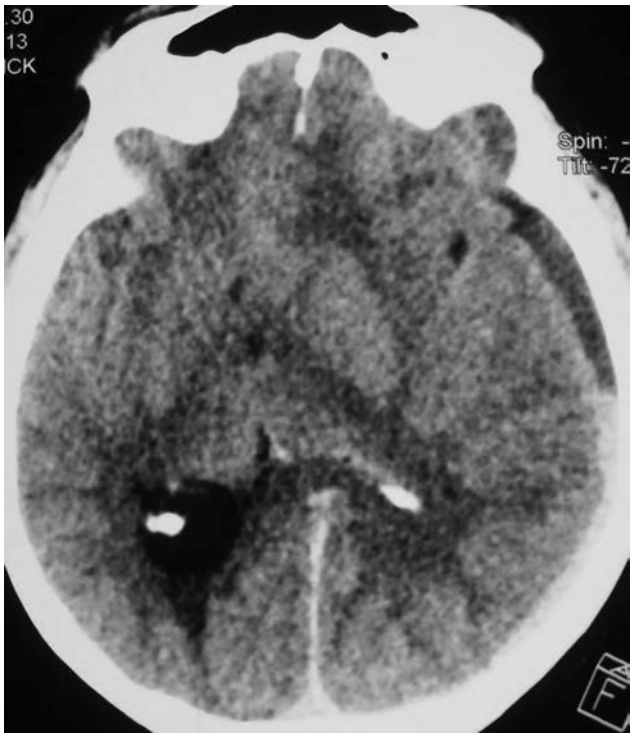

Fig. 2 Axial head computed tomography without contrast medium showing a mass effect, double-density chronic subdural hematoma located in the left cerebral hemisphere.

Discussion

Hemiparesis ipsilateral to a brain injury is called Kernohan-Woltman[6] notch sign, and it is considered a false localizing neurological sign. The Kernohan-Woltman phenomenon was initially described by Kernohan et al in 1928, in a necropsy examination of a brain tumor patient with a notched brain peduncle from a contralateral herniation.[8] The condition has been associated with brain tumors and supratentorial-located hematomas.[5]

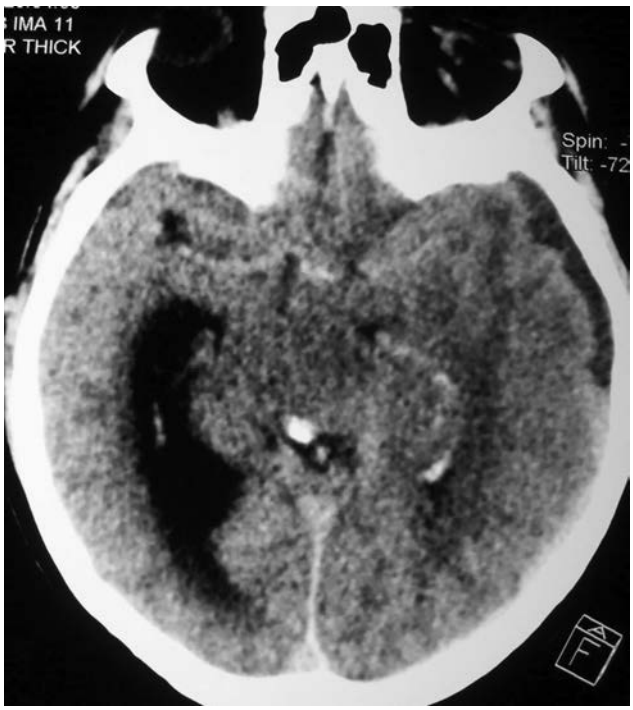

Fig. 3 Axial head computed tomography without contrast medium showing a chronic subdural hematoma in the left cerebral hemisphere and obliteration of the homolateral ventricle with obliteration of the interpeduncular cistern.

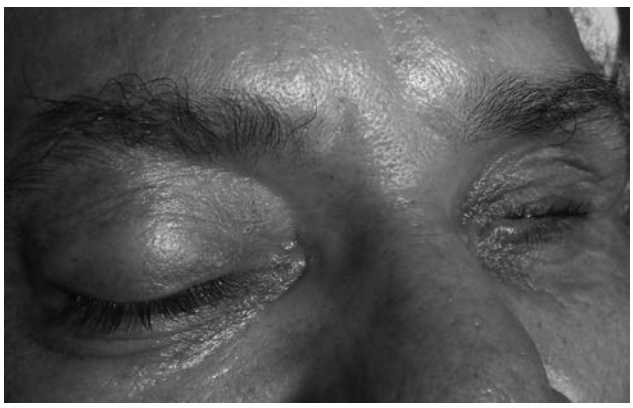

Fig. 1 Absence of the left eye (enophthalmos).

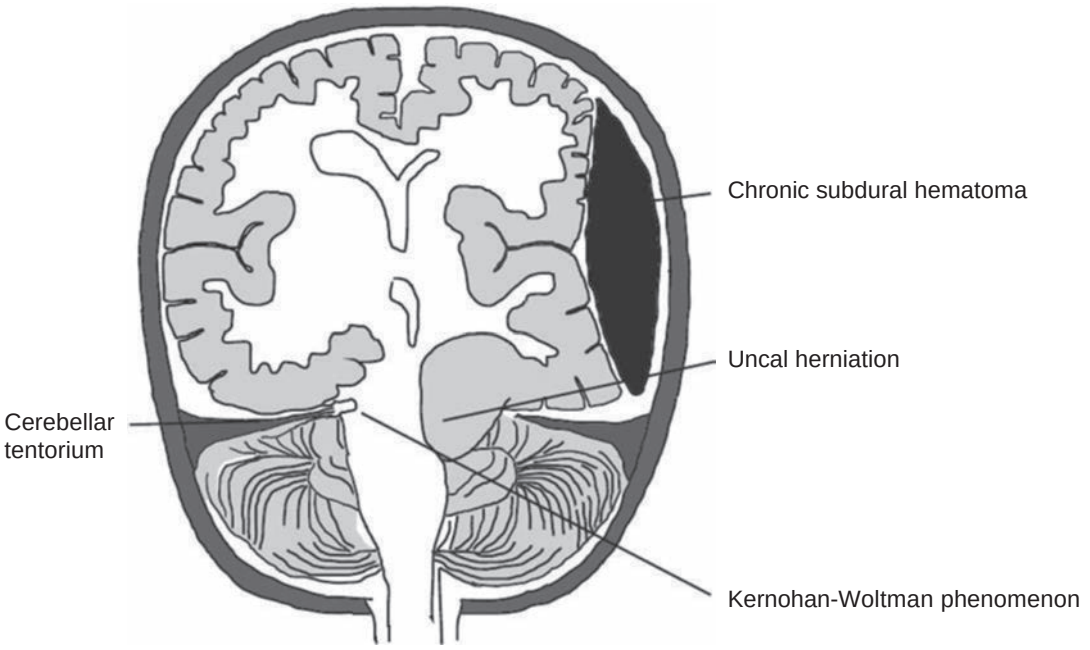

Fig. 4 Schematic representation of the Kernohan-Woltman notch phenomenon.

Transtentorial herniation is a neurological phenomenon well described in the medical literature, and it is associated with a supratentorial expansive lesion. This herniation causes direct neural compression and indirect effects of vascular compromise, leading to obstructive hydrocephalus and herniation of the strangled tissue.[1]

Uncal herniation is a common subtype of transtentorial herniation, in which the most internal part of the temporal lobe, the uncus, can be compressed against the tentorium, forming a recess at the crus cerebri known as Kernohan-Woltman notch and compressing the brainstem, mainly the midbrain.[2,5,8] With time, the integrity of the crus cerebri (the frontal aspect of the cerebral peduncle) and of the corticospinal descending tract is compromised, causing contralateral motor deficit associated with a decreased level of consciousness.

The Kernohan-Woltman notch signal is a rare condition in which the peduncle contralateral to a supratentorial expansive lesion is forced against the end of the tentorium, leading to hemiparesis ipsilateral to the lesion (►**Fig. 4**). The rigid edge of the tentorium can transect the brain peduncle, particularly the fibers protruding to the leg. The lesion can occur in the absence of an uncal herniation, probably due to a rapid acceleration-deacceleration leading to a backlash injury of the cerebral peduncle.[1] Therefore, the hemiparesis ipsilateral to a supratentorial expansive lesion is known as Kernohan-Woltman phenomenon. This phenomenon is considered a false localizing neurological sign.

The diagnosis of the Kernohan-Woltman phenomenon is based on neurological examination and on neuroimaging findings.[2,8] The neurological condition is characterized by mydriasis and hemiparesis, and it may be associated with a decreased level of consciousness.[4]

A CT scan can demonstrate the effect of an expansive mass lesion compressing the brainstem, while the identification of an uncal herniation is more difficult.[5] A resonance magnetic imaging (MRI) exam has better definition in multiple planes and better resolution to evaluate the brain stem, being able to reveal a deformity or injury in the cerebral peduncle resulting from a transtentorial herniation.[8–10] The Kernohan-Woltman phenomenon has been identified in coronal T2-weighted and fluid attenuation inversion recovery (FLAIR) sequences, and it tends to present a peripheral triangular morphology.[1,8] According to Moon et al,[11] the lesion is a small, hypointense signal in T1-weighted images, with a hyperintense signal in T2-weighted images, in the anterolateral region of the midbrain.

In the present case, due to the clinical conditions of the patient in the emergency room, only one CT scan was performed, but the neurological picture was consistent with the Kernohan-Woltman notch signal.

Patients who present with the Kernohan-Woltman notch phenomenon are surgically treated; however, in severe cases, hyperventilation, administration of osmotic diuretics, and general care can be performed, not always with a good prognosis.

The Kernohan-Woltman notch signal has been considered a rare finding. Magnetic resonance imaging scans have been used to confirm the diagnosis. The present case serves as a warning for emergency physicians to be aware of its severity and of the fact that it may not always be possible to identify this phenomenon in CT scans as a false localizing neurological sign in order to avoid an incorrect surgical approach.

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Clival Subdural Hematoma after Drainage of Concomitant Intracranial and Spinal Cord Subdural Hematomas – Rare Case Report

Hematoma subdural de clivo após drenagem de hematomas subdural intracraniano e medular concomitantes – raro relato de caso

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Abstract

Concomitant traumatic spinal cord and intracranial subdural hematomas associated with a retroclival hematoma are very uncommon. Their pathophysiology is not totally elucidated, but one hypothesis is the migration of the hematoma from the head to the spine. In the present case report, the authors describe the case of a 51-year-old man presenting with headache, nauseas and back pain after a head trauma who presented with intracranial and spinal cord subdural hematomas. Drainage was performed but, 1 week later, a retroclival subdural hematoma was diagnosed. The present paper discusses the pathophysiology, the clinical presentation, as well as the complications of concomitant traumatic spinal cord and intracranial subdural hematomas associated with a retroclival hematoma, and reviews this condition.

Keywords
- lumbar subdural hematoma
- intracranial subdural hematoma
- retroclival subdural hematoma
- head trauma

Resumo

Hematomas subdurais traumáticos medular e intracraniano concomitantes associados a hematomas retroclivais são condições incomuns. Sua fisiopatologia não é completamente compreendida, mas uma das hipóteses é a migração do hematoma da região encefálica para a medular. No presente relato de caso, os autores descrevem o caso de um paciente masculino, 51 anos de idade, com cefaleia, náuseas e dor lombar após sofrer traumatismo craniano. O mesmo apresentou-se com hematomas subdurais encefálico e lombar. O paciente foi submetido a drenagem e, após uma semana, retornou com um hematoma subdural retroclival. O presente artigo discute a fisiopatologia, a apresentação clínica e as complicações de hematomas subdurais traumáticos medular e intracraniano concomitantes associados a hematomas retroclivais e apresenta uma revisão do tema.

Palavras-Chave
- hematoma subdural lombar
- hematoma subdural intracraniano
- hematoma subdural retroclival
- traumatismo craniano

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Introduction

Concomitant traumatic spinal cord and intracranial subdural hematomas are unusual, and even rarer when associated to a late retroclival subdural hematoma.

Unlike the intracranial subdural space, the spinal cord subdural content has no bridged veins; as such, the hypothesis of rupture of vessels in this region is less plausible, and the hypothesis of migration of the intracranial hematoma to the lumbar region seems more likely.

Other possible causes for this condition include coagulation disorders, lumbar puncture, anticoagulation therapy, and idiopathic causes.

Most cases of traumatic hematomas in the posterior fossa are extradural and occur in children who suffered head injury due to automobile accidents. Its physiopathology can be associated to ruptures of ligaments or to bone fractures at the base of the skull.

The present case report describes a case of chronic spinal cord and intracranial subdural hematoma, subsequently associated with a clival subdural hematoma.

Case Report

Male patient, 51-years-old, with head trauma due to a wood log that fell on the right parietal region ~ 20 days before admission. The patient reported progressive headache accompanied by nausea and back pain, walking difficulties, and no improvement with common painkillers. At the physical examination, the patient presented grade IV strength in the right leg, in addition to a positive Lasègue sign on the same limb. There was no history of

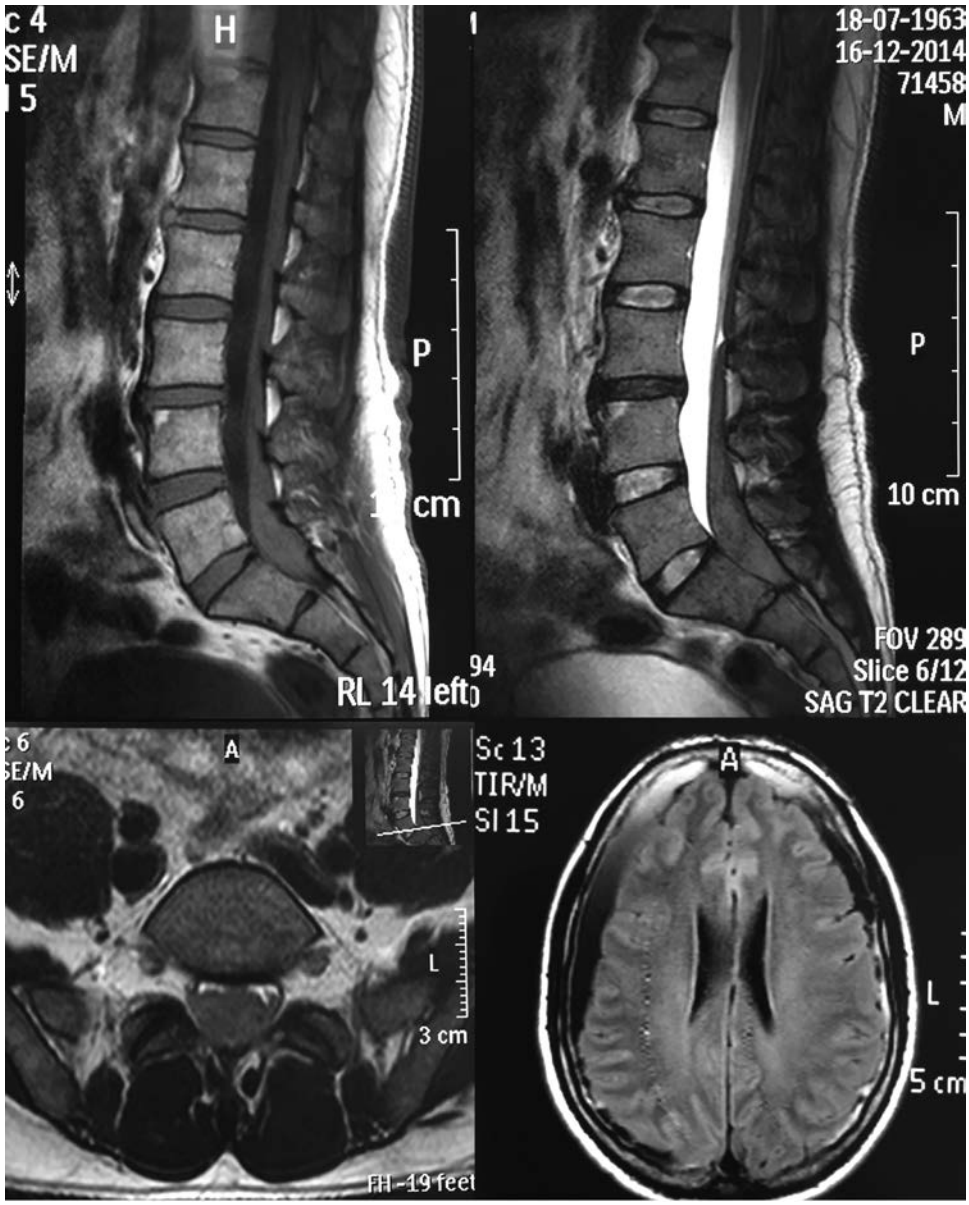

Fig. 1 (A and B) T1 and T2-weighted magnetic resonance imaging of the lumbar spine showing a subdural hematoma in L3-S2. (C) Axial T1-weighted sequence showing the lumbar subdural hematoma. (D) FLAIR sequence revealing a bilateral brain subdural hematoma. Abbreviations: FLAIR, fluid attenuation inversion recovery.

coagulopathies, of comorbidities or of use of medications. A magnetic resonance imaging (MRI) of the brain and of the lumbar spine (►**Fig. 1**) showed a bilateral brain and spinal cord injury with extension from the L3 to the S2 vertebrae that was isointense at T1 and T2-weighted sequences and compatible with bilateral intracranial and spinal cord subdural hematomas.

Test results for coagulopathies were within the normal limits.

The patient was then submitted to drainage of the spinal cord hemorrhage through a hemilaminectomy from the L4 to the S1 vertebrae, followed by a durotomy (►**Fig. 2**). In the same surgery, drainage of the intracranial subdural hematoma was performed by a trepanation in each hemisphere. Since the patient evolved stably, and the clinical picture improved after the surgery, he was discharged 4 days later. One week after the discharge, the patient complained of recurrence of the headache, and a new magnetic resonance imaging (MRI) of the brain showed a clival subdural hematoma, which we decided to follow-up in an outpatient facility.

Thirty days later, the patient came for an outpatient visit and presented improvement of the headache and of the motor deficit in the right lower limb.

Subsequently, the MRI of the brain was repeated and showed the absorption of the retroclival subdural hematoma.

Discussion

Lumbar subdural hematoma is an uncommon condition found in $< 5\%$ of the cases of lumbar hematomas in retrospective studies. The extradural hematoma is more common and was presented in $\sim 74\%$ of the patients.[1,2]

The present case had no coagulation disorders, had not been submitted to a recent anesthetic procedure, and had no history of lumbar puncture, although these are common associations, reaching 30% of the iatrogenic causes of lumbar subdural hematoma.[3,4]

The association between intracranial and spinal cord subdural hematomas is rare, with 17 cases in the literature, according to our survey.

Kokubo et al performed a prospective study with 168 patients with chronic subdural hematoma with surgical indication between August 2007 and September 2011. These patients were screened for lumbar subdural hematoma by an MRI of the lumbar spine, which revealed 2 cases with concurrent hematomas (1.2%).[5,6]

Our patient reported head trauma and denied any spinal trauma or falling from his own height, which seems to corroborate the hypothesis of migration of the cranial subdural hematoma to the lumbar region, as proposed by Bortolotti et al This would be possible due to the influx of

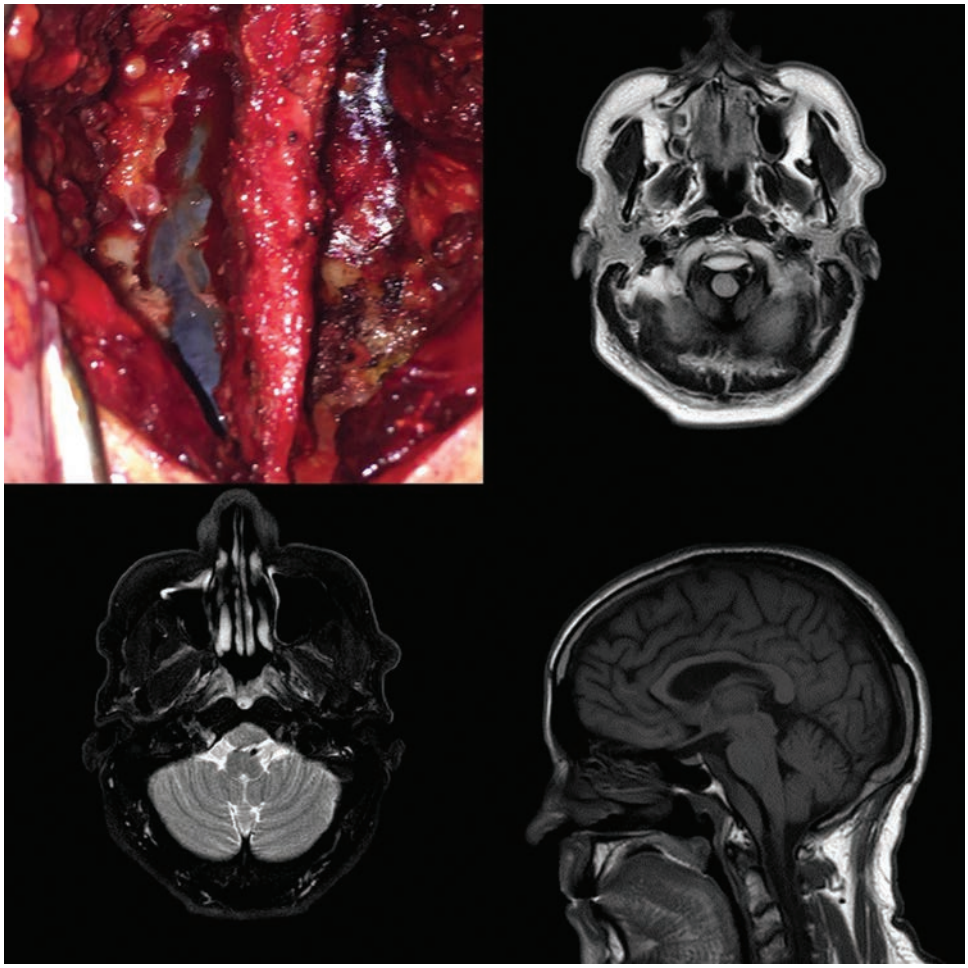

Fig. 2 (A) Intraoperative image of hematoma drainage. (B, C, and D) Axial FLAIR sequence, T1-weighted and sagittal T2-weighted magnetic resonance images of a retroclival subdural hematoma. Abbreviations: FLAIR, fluid attenuation inversion recovery.

cerebrospinal fluid (CSF) through the subdural space, diluting the hematoma and facilitating its migration. Moreover, according to these authors, the formation of subdural hematomas at the spinal level would be difficulted by the avascular plane of the subdural space at this site.[7,8]

Hung et al suggested that the higher intracranial pressure increases the shear forces between the dura mater and the arachnoid mater, creating a space in which the hematoma can progress to the region of the spinal cord.[9–11]

Spinal cord subdural hematomas associated with deficit can be treated by surgical drainage with laminectomy and durotomy as quickly as possible, as these procedures improve the evolution of the case, leading to recovery in 80% of the cases.[12]

Traumatic retroclival hematomas are mostly extradural, secondary to traffic accidents, and are more frequent in the pediatric population, presenting clinically as abducens nerve palsy. Retroclival subdural hematomas, on the other hand, account for 0.3% of all subdural hematomas. To our knowledge, there are rare reports of traumatic origin in adults, who, like in the present case, did not present with any motor deficits and evolved benignly.[13,14]

The physiopathology of concomitant traumatic spinal cord and intracranial subdural hematomas associated with a retroclival hematoma remains uncertain, but, as in clival extradural hematomas, it is hypothesized that these hematomas arise from a fracture or ligament rupture associated with a venous lesion.

We believe that the drainage of the spinal cord hematoma caused the migration of part of the intracranial hematoma to the clival region, leading to the late onset of headache.[15,16]

The treatment of clival subdural hematomas varies according to their clinical presentation; conservative therapies are preferred in cases with no deficits.[17,18]

Conclusion

The association of cerebral, spinal and clival subdural hematomas is uncommon in the literature. Due to its rarity and to the lack of specificity of most of its signs and symptoms, this combination can be misdiagnosed. Its treatment depends on the clinical picture of the patient; in cases with motor deficits, surgical therapy provides a better prognosis.

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Tuberculoid Granuloma in the Brainstem: Case Report

Granuloma tuberculoide no tronco cerebral: relato de caso

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Abstract

Meningitis or meningoencephalitis are the most common presentations of Koch bacilli infection on the central nervous system (CNS), especially in immunosuppressed patients, in whom the bacilli normally reaches the meninges and the cerebral parenchyma.. A least common pathological presentation is the tumoral growth pattern disease known as tuberculoma. This pathological entity is more common in the cerebral hemispheres and is rarely located in the brainstem. The present case report describes a case of a 55-year-old patient under regular antiretroviral therapy who was hospitalized with signs of brainstem and cerebellar disturbances. Computed tomography (CT) and magnetic resonance imaging (MRI) of the brain showed an exophytic lesion in the dorsal region of the pons. The patient underwent total resection of the lesion and the histopathologic analysis was consistent with a tuberculoma.

Keywords
- tuberculoma
- granulomatous tuberculosis
- brainstem

Resumo

O acometimento do sistema nervoso central pelo bacilo de Koch é comum, principalmente em pacientes imunossupressos, nos quais normalmente atinge as meninges e o parênquima cerebral, causando quadro de meningite ou meningoencefalite. Contudo, menos comumente, tal acometimento pode formar lesões expansivas conhecidas como tuberculomas. A presença deste tipo de tumoração predomina no parênquima supratentorial, sendo rara sua localização no tronco cerebral. O presente relato de caso tem por objetivo descrever o caso de um paciente de 55 anos, em uso de terapia antirretroviral, que apresentou inicialmente síndrome piramidal alterna com componentes cerebelares. Tomografia e ressonância magnética de crânio mostraram a presença de lesão expansiva exofítica em região dorsal da ponte, no assoalho do quarto ventrículo. O paciente foi submetido à ressecção total da lesão, e o exame anatomopatológico foi compatível com tuberculoma.

Palavras-chave
- tuberculoma
- granuloma tuberculoide
- tronco cerebral

Introduction

The World Health Organization (WHO) estimates that a third of the population of the world is infected with *Mycobacterium tuberculosis*; people coinfected with HIV are at a greater risk of developing the active and widespread form of the disease, including central nervous system (CNS) involvement.[1] However, only 50% of the cases of neurotuberculosis are associated with active extraneural

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tuberculosis, and its incidence is increasing in people < 20 years old.[2]

Clinically, the most common manifestation is neurotuberculosis, meningeal disease, but parenchymatous involvement secondary to tuberculomas, tuberculous abscesses, focal cerebritis or tuberculous allergic encephalopathy are also possible.[2–7]

To broaden the understanding of the diagnosis and of the treatment of brainstem tuberculomas, the present case report describes a case and discusses its possible differential diagnoses.

Case Report

Male patient, 55 years old, presenting with right-sided paresthesia, ataxia and diplopia for 1 week. During the anamnesis, the patient reported positivity to human immunodeficiency virus (HIV) and to hepatitis C virus (HCV) and antiretroviral therapy use (efavirenz, lamivudine and zidovudine). His treatment was followed-up by an infectious disease specialist, and tests performed shortly before hospitalization showed a CD4 count of 387 and a viral load of log 2.9. The patient was alert, with adequate level and content of consciousness. The physical examination of the cranial nerves showed internuclear ophthalmoplegia and left-sided conjugate gaze palsy. He presented with facial sensitivity with clear right-sided facial hypoesthesia and left peripheral facial paralysis, but with no disfiguration. In addition, the patient had central hypoacusis. Cerebellar tests were consistent with right upper limb dysmetria, but with no dysdiadochokinesia, no hypotonia and no altered results in the Stewart-Holmes test. Finally, the patient had no change in tactile, thermal, vibration, painful or appendicular proprioceptive sensitivity, but presented with a disproportionate, right-sided grade IV hemiparesis with distal brachiocrural predominance. The alternating syndrome with cerebellar syndrome components led to the diagnosis of a brainstem lesion with possible direct or indirect compromise of the right cerebellar hemisphere.

The investigation continued with imaging tests. A computed tomography (CT) of the head showed a hypodense lesion in the posterior fossa with compression of the fourth ventricle, in the pons and midbrain topography, predominantly left-sided. A magnetic resonance imaging (MRI) of the skull showed a lobular expansive lesion in the dorsal portion of the pons, measuring 2.2 × 2.1 × 2.0 cm. This lesion was hypointense in T1-weighted images and discretely hyperintense in T2-weighted images, also presenting a heterogenic enhancement after gadolinium infusion and a vasogenic edema extending to the bulb, to the left cerebellar hemisphere and to the mesencephalon (►**Figs. 1**, **2** and **3**). Since the patient was immunosuppressed, a fungal opportunistic infection was suspected, and, following standardized flow charts to investigate this type of lesion, a CT of the chest was also performed in search for a probable underlying infectious site, which revealed a 4 mm nodule in the right lower lobe and bilateral apical centrilobular micronodules, with no signs of active disease, pulmonary abscesses or "fungal balls." A bronchoalveolar lavage was also performed and it was negative for Koch bacilli and fungi (both in the direct examination and in cultures).

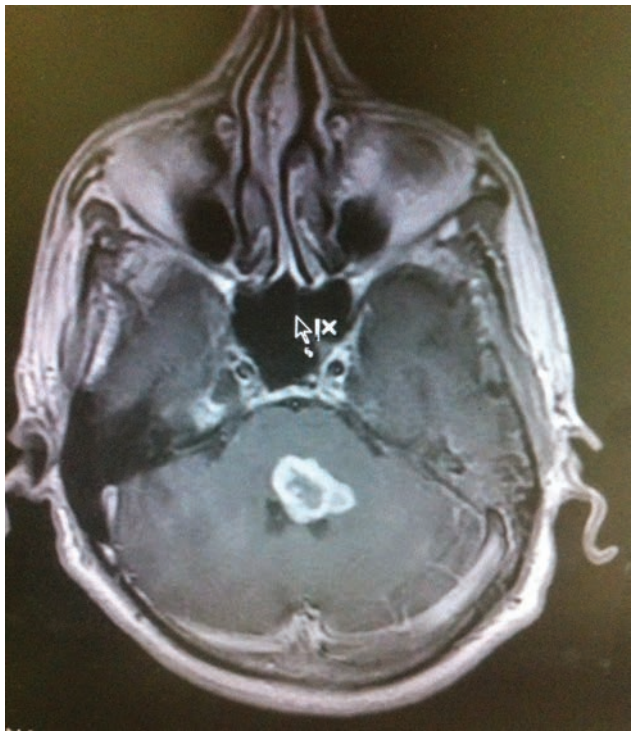

Fig. 1 Axial magnetic resonance imaging enhanced with gadolinium showing an irregular outlined lesion with an isointense center circumscribed by a hypersignal area with contrast medium uptake located in the posterior portion of the pons and in the left middle cerebellar peduncle, and an exophytic growth toward the fourth ventricle.

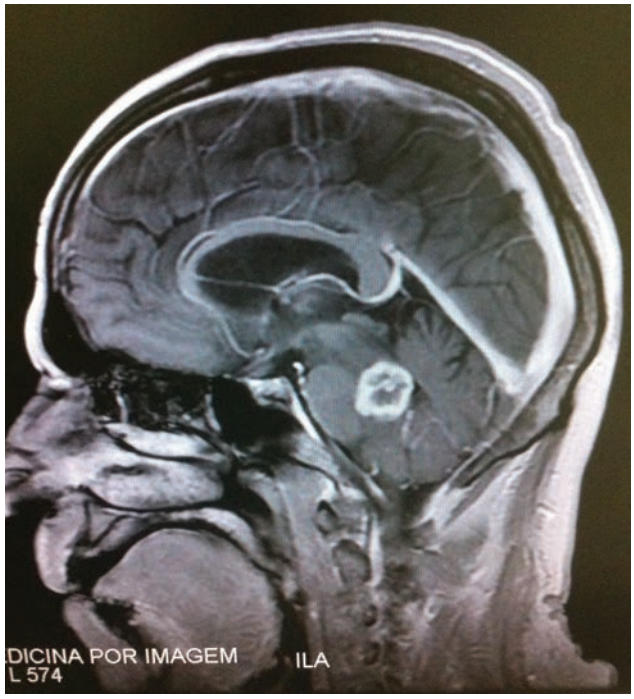

Fig. 2 Sagittal magnetic resonance imaging enhanced with gadolinium showing a lesion with peripheral contrast medium uptake in the posterior portion of the pons and growing toward the fourth ventricle.

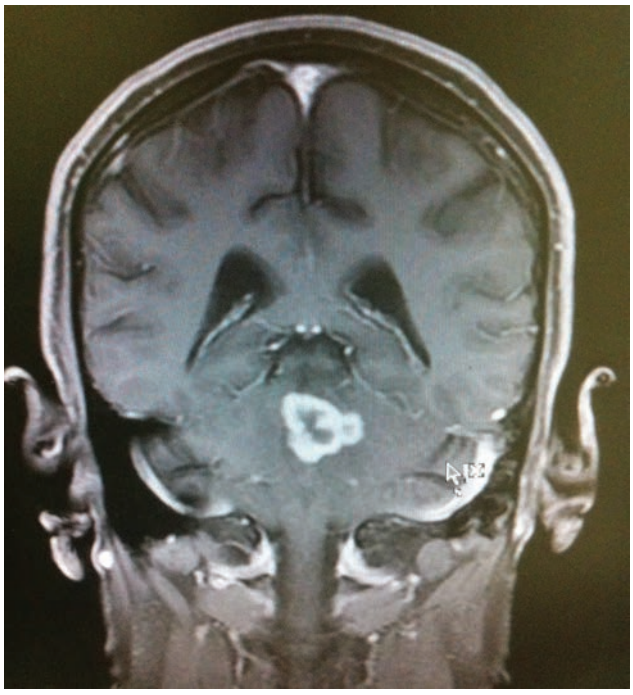

Fig. 3 Coronal magnetic resonance imaging enhanced with gadolinium showing a lesion with peripheral contrast medium uptake and content with intensity similar to the cerebral parenchyma.

With no diagnosis, the patient was submitted to a neurosurgical approach through a telovelar access. Intraoperative visualization aided by a microscope allowed the identification of a red-colored, apparently infiltrative, exophytic lesion. An incisional biopsy of the lesion was attempted, but it was unsuccessful because the lesion was very hardened. A cleavage plane between the lesion and the parenchyma was then identified and dissected, allowing the complete resection of the diseased tissue. After its removal, a macroscopic analysis of the lesion revealed a granulomatous appearance. The histopathologic examination was consistent with a tuberculoid granuloma.

In the early postoperative period, the patient continued to present with the previous deficits and evolved with a worsening of the left facial paralysis (with disfiguring asymmetry) and right palpebral ptosis. These findings improved progressively, and the patient presented good clinical and neurological evolution. On the 10th postoperative day, the patient presented with a sudden reduction of the level of consciousness and anisocoria (with left-sided mydriasis). An urgent CT of the head identified a massive acute subdural hematoma. Although an urgent surgery for subdural hematoma drainage was indicated, the patient died within 2 days.

Discussion

Approximately one third of the population of the world is infected with *M. tuberculosis*, and its coinfection with HIV can be the most important factor for systemic infection.[1] The number of coinfected people is estimated in 3.1 million.[8] In Brazil, the annual tuberculosis incidence dropped from 51.8 to 38.2% in the last 2 decades. In addition, there was a 26% reduction in the incidence and a 32% reduction in the general mortality due to tuberculosis.[9]

The most common form of tuberculosis of the CNS is tuberculous meningoencephalitis,[2] followed by expansive focal lesions, mostly supratentorial, with few reported cases involving the brainstem and the cerebellum.[2–7]

Among the CNS infections by *M. tuberculosis,* tuberculomas can account for between 5 and 30% of all of the expansive brain lesions; however, brainstem lesions remain uncommon, corresponding to between 2.5 and 8% of the cases.[3,5,6]

In the brain, granulomas or Rich foci can form between the subpial and subependymal layers, and expand to create tuberculomas or abscesses at the cerebral parenchyma; or, more commonly, they break up, causing meningitis.[1,10] These lesions are more frequent at the corticomedullary junction, especially in the frontal and parietal lobes, due to the hematogenous dissemination of miliary tuberculosis.[2] The most common clinical manifestation of tuberculomas is headache, as well as signs of intracranial hypertension and focal neurological deficit.[2–4] Brain abscesses rarely cause fever and alteration in the level of consciousness, but $>$ 25% of the patients have convulsive crises.[11]

Tuberculomas commonly present as well-delimited, nodular, hardened and avascular lesions, involved by edema and gliosis areas.[7] At MRI scans, these lesions appear as hypointense or isointense in T1-weighted images, but their appearance in T2-weighted images is different according to their pattern: caseous lesions present a ring pattern with central necrosis; solid lesions show homogeneous enhancement.[2] There is also a third manifestation, rarer than those previously mentioned, in which the lesion is associated with cysts.[7]

Rapid detection in HIV coinfected patients is crucial, but finding acid-fast bacilli by polymerase chain reaction (PCR) or positive culture is rare.[12] Neither the tuberculin test (31% sensitivity) nor the interferon gamma detection blood test (IGRA test) (60% sensitivity) can exclude with precision tuberculosis in HIV-positive individuals, especially if CD4 counts are $<$ 200 cells/µl.[13] Repeated, high-volume lumbar punctures improve the yield of these tests.[14]

The treatment for tuberculosis does not differ significantly in patients coinfected or not with HIV.[15] The gold standard is the initial institution of isoniazid, pyrazinamide, ethambutol and rifampicin for 2 months.[11] Rifampicin decreases protease inhibitors and nevirapine plasma levels, and rifabutin is an appropriate alternative.[16] After the initial phase, isoniazid and rifampicin or rifabutin are continued for between 9 and 12 months.[15] The use of steroids and the ideal time to start antiretroviral therapy (ART) along with antituberculosis therapy remain controversial.[15]

Although CNS tuberculosis is clinically managed, there are some surgical indications, either for the diagnosis (as in the present case) or for the treatment of the granuloma or of infectious complications, such as the need for ventricular shunts due to changes in cerebrospinal fluid (CSF) dynamics.[3–5,8]

Neurosurgery is imperative for the identification of the causative organism if it has not been otherwise determined and, in selected cases, to reduce the size of the abscess.[11] Stereotaxic surgery allows the aspiration of virtually any

abscess with at least 1 cm in diameter regardless of its location; the aim of the diagnosis should be the maximum possible drainage.[11] Moreover, it also aims decompression, unless there is a contraindication due to the suspected organism or in relation to the clinical condition of the patient.[11]

A neurosurgical intervention is recommended in abscesses $>$ 2.5 cm in diameter.[17] However, data from comparative studies are limited, and this size cannot be regarded as a definite indication for aspiration.[11] In patients with multiple abscesses, the larger abscesses must be aspirated to ascertain the diagnosis.[11]

In the past 50 years, the prognosis has progressed due to new testing techniques, to antimicrobial therapy regimens, and to the introduction of minimally invasive neurosurgical procedures.[18] However, mortality in multidrug-resistant HIV-coinfected patients remains extremely high.[19]

Conclusion

Brainstem tuberculomas can be systemically treated with specific medications and surgically managed in case of failure to respond to the medical treatment. However, the surgical removal of expansive brainstem lesions can be recommended in cases in which the lesion is related to the floor of the fourth ventricle, since this approach is feasible and capable of obtaining good results in the postoperative period.

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Resting Tremor after Mild Head Injury: Case Report

Tremor de repouso após traumatismo cranioencefálico leve: relato de caso

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Abstract

Over the past few decades, it has been recognized that traumatic brain injury (TBI) may result in various movement disorders. However, moderate or mild TBI only rarely causes persistent post-traumatic movement disorders. In the present report, we describe a case of secondary tremor due to a mild head injury with a transitory loss of consciousness. A 26-year-old man developed an isolated rest tremor of the hands and legs without other neurologic signs. The interval between the head trauma and the onset of the symptoms was 4 months. Neuroimaging studies reveled gliosis in the lentiform nucleus. Haloperidol administration resulted in tremor reduction. A rest tremor, similar to essential tremor, can be a rare complication of head trauma. Haloperidol may be an effective and safe treatment modality for post-traumatic tremor. Further studies are needed to clarify the optimal drug for the treatment of post-traumatic tremor.

Keywords
- traumatic brain injury
- movement disorder
- posttraumatic
- tremor

Resumo

Nas últimas décadas, tem-se reconhecido que o traumatismo cranioencefálico (TCE) pode resultar em diferentes desordens de movimento. No entanto, somente em raros casos o TCE leve e moderado pode desencadear desordens de movimento. O presente relato apresenta um caso de tremor secundário a um TCE leve, com perda transitória da consciência, cujo paciente, com 26 anos de idade, do sexo masculino, desenvolveu um quadro neurológico de tremor de repouso das mãos e pernas de forma isolada, sem outros sinais neurológicos associados. Note-se que o intervalo entre o TCE e o início dos sintomas foi de 4 meses. O estudo de neuroimagem revelou gliose bilateral no núcleo lentiforme. A administração de haloperidol resultou em controle dos sintomas. Tremor de repouso similar ao tremor essencial é uma rara complicação do TCE. Dentre as modalidades de tratamento efetivas e seguras para o tremor pós-TCE, encontra-se o haloperidol. Entretanto, mais estudos são necessários para definir o medicamento que se mostra mais adequado para o tratamento do tremor pós-TCE.

Palavras-Chave
- traumatismo cranioencefálico
- desordem de movimento
- pós-traumático
- tremor

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Introduction

Traumatic brain injury (TBI) is a cerebral insult caused by an external physical force promoting an anatomical lesion and/or functional compromise of the scalp, of the cranium, of the meninges, or of the brain.[1,2] Traumatic brain injury is one of the major global public health problems, with a high and increasing incidence in the modern world.[1,2] It also represents an important cause of morbimortality in adolescents and young adults, leading to a decrease in the productive capacity and in societal financial losses.[1,2] Regardless of the socioeconomical status, the number of deaths resulting from TBI is surpassed only by neoplastic and cardiovascular diseases.[2]

The incidence of TBI in the United States was estimated in 538.2 cases per 100,000 people; in Europe, in 235 cases per 100,000; and in Australia, in 322 cases per 100,000.[3] In Brazil, annually, around half a million people are hospitalized due to TBIs.[2] In the past 10 years, more than 1 million people became disabled due to traffic accidents.[1]

The mechanism of TBI can be divided in: (1) focal brain lesion, resulting in contusion, laceration, and intracranial hemorrhage due to direct local trauma; and (2) diffuse brain lesion, which causes diffuse axonal lesion and brain swelling (edema) by the mechanism of acceleration-deacceleration.[4] The outcome of the brain lesion is defined by two different mechanisms: (1) primary lesion, which occurs at the moment of the trauma; and (2) secondary lesion, resulting from late clinical manifestations of the pathological process started by the trauma.[4]

The disabilities resulting from TBI can be divided into three categories: physical (sensory-motor), cognitive, and emotional/behavioral.[5] Physical disabilities are diversified and may include motor, visual, and tactile disabilities. Cognitive disabilities often include attention, memory, and executive function disorders.[5,6]

Post-TBI extrapyramidal alterations are rare, and their manifestation can overlap with Parkinsonism (tremor, dyskinesia and rigidity), which are considered hypokinetic alterations, or with hyperkinetic alterations (chorea, ballismus, and athetosis).[7–9]

The present report describes the case of a patient with post-TBI rest tremor, a rare sequela in this scenario.

Case Report

Patient W. A. C., male, 26 years old, civil construction worker, married with no children. While riding a bicycle, the patient collided with a motorcycle. The patient lost consciousness at the site of the accident and was transferred to a reference hospital by the mobile emergency service. After the initial treatment, the following tests were performed and did not reveal any abnormalities: computed tomography (CT) of the head, cervical and lumbar spine X-rays, and chest X-ray. Since the patient scored 14 points in the Glasgow coma scale at admission, he remained at the hospital for a 24-hour period.

Four months later, the patient presented to the medical office complaining of rest tremors in the limbs and gait alteration, requiring time off work. In order to perform a more thorough investigation, a magnetic resonance imaging (MRI) exam of the head was performed and revealed small foci of gliosis at the left putamen and at the globus pallidus bilaterally (►**Fig. 1**). The patient did not report any neurological conditions in his previous or familial history. At the neurological exam, he did not present cranial nerves or muscle strength abnormalities, neither dysmetria nor nystagmus. A frequency rest tremor, more evident in the inferior limbs, mainly on the left side, and of mild intensity in the superior limbs, was observed. Gait was impaired by the tremors in the inferior limbs. Data collected at anamnesis, at the physical exam, and at the neurological exam identified a condition consistent with post-severe TBI hyperkinetic extrapyramidal syndrome, of the rest tremor type. Treatment with haloperidol 2.5 mg was initiated and controlled the symptoms.

Discussion

Traumatic brain injury is more frequent in young adults and adolescents.[1] Its incidence varies according to gender, being higher in men than in women, possibly reflecting differences in the exposure to risk situations.[1] Regarding the external causes of trauma, accidents with motor vehicles—cars and motorcycles—have been largely responsible for TBI cases.[2,3] The most commonly injured cranial area in TBI is the frontal region; intracranial hemorrhage is more common than traumatic subarachnoid hemorrhage (TSAH).[1,2]

A definite brain lesion established after a TBI results from physiopathological mechanisms initiated by the accident, which may last for days or weeks.[10] Acceleration, deacceleration, and rotational forces, as well as piercing objects, can cause laceration, compression, tension, or shearing of tissues, or even a combination of such harmful events, which result in primary brain lesions – concussion, brain contusion, or diffuse axonal lesion of the white matter.[10] Causes for secondary lesions can arise during the trauma or after some time.[11,12]

The cognitive sequelae observed in TBI patients include somatic, perceptive, cognitive, and emotional symptoms that can be characterized as a postconcussion or as a posttraumatic syndrome.[11] This condition has been described more frequently in patients who did not require hospitalization or who had brief post-traumatic amnesia, lasting for < 24 hours, and who did not present with any abnormalities in neuroimaging scans.[11]

Regarding motor sequelae, many patients present a hemiplegic and/or hemiparetic pattern, with gait alterations and functional mobility abnormalities in the superior and/or in the inferior limbs due to lesions in the pyramidal tract.[5] Lesions involving the pyramidal tract evolve with spasticity and exacerbated tendinous reflexes.[13]

Involuntary movements that can compromise voluntary motricity may be observed if the lesions involve the basal ganglia.[13] The frequency of involuntary movements as a sequela of TBI is not well known due to the limited number of publications in the medical literature.[14–17] In the pediatric group, tremor as a sequela of TBI was observed in up to 45% of the patients, and the symptoms began, in $\sim$ 90% of the cases, up to 12 months after the trauma.[18]

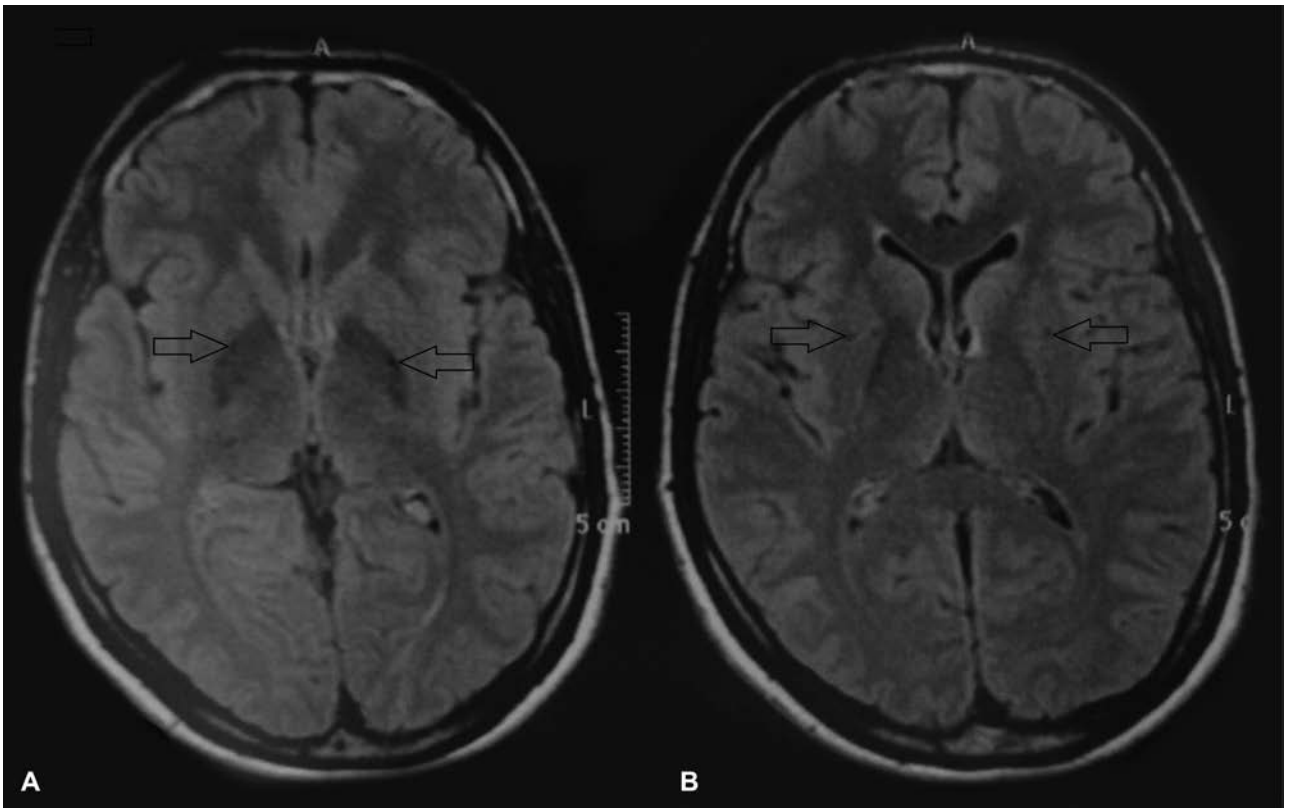

Fig. 1 Cranial axial magnetic resonance imaging – Flair sequence. (**A**) Note the presence of bilateral hypointense points, indicated by the arrows, in the globus pallidus, corresponding to areas of post-traumatic brain injury gliosis. (**B**) Note the presence of bilateral hypointense points, indicated by the arrows, in the putamen, also corresponding to areas of post-traumatic brain injury gliosis.

Extrapyramidal syndromes can be classified in two categories: hypokinetic syndromes (akinetic-rigid), and hyperkinetic syndromes.[8,13] The former are clinically characterized by a reduced velocity in spontaneous movements (bradykinesia) and rigidity.[8] The latter are characterized by abnormal involuntary movements (tremor, chorea, athetosis, and ballismus).[8]

According to the phenomenological classification by Jankovic, tremors can be divided in rest tremors, action tremors, miscellaneous tremors, and other rhythmic movements.[19]

- Rest tremor is characterized as a limb tremor corresponding to the muscles that were not activated for the movement.[20]
- Action tremor occurs when there is a voluntary muscle contraction. It can be subdivided in postural, kinetic, positional, task-specific, and isometric tremors.[19]
- Postural tremor occurs when moving to the orthostatic position in order to move towards an object.[21]
- Kinetic tremor occurs when movement is initiated, and it is sustained for the duration of the movement.[21]
- Position-specific tremor occurs in certain postures; for instance, with a cup or glass near the mouth, or when in biped position; in the latter case, it is called orthostatic tremor.[21]
- Action-specific tremor occurs in certain activities; for instance, writing (primary writing tremor), speaking or singing (vocal tremor).[21]
- Isometric tremor appears during voluntary muscle contraction sustained against a certain fixed object.[21]
- Essential tremor is the most common pathological tremor and the most frequent movement disorder.[21,22] It is classically defined by a bilateral, fine postural tremor, visible to the naked eye, persistently affecting the hands, lasting for $>$ 5 years.[21,22] Moreover, it can be associated with a kinetic tremor or with a concomitant tremor in other body regions, as in the hands. Sometimes, the amplitude of the tremor fluctuates, often between 4 and 12 Hz.[21,22]
- Fine tremor affects the ability to control small muscles for delicate and specific movements, such as in writing.[23]
- Gross tremor involves wide movements, such as walking and running, through the contraction of large body muscles.[23]

There is also a tremor classification considering its electrophysiological data: low frequency tremors present in frequencies between 2.5 and 4 Hz, and high frequency tremors present in frequencies $>$ 5 Hz.[19]

It should be emphasized that the pathophysiology of the extrapyramidal syndrome due to TBI is independent from the severity and presence of coma, although the neuroanatomic basis for the post-traumatic tremor remains unknown.[20] It has been observed that the postural and kinetic tremor of the hand is often asymmetrical and tends to be concomitant to head, leg and trunk tremors, beginning several weeks after the cranial injury.[20]

A study by Johnson et al suggested that midbrain lesions could be associated with the genesis of extrapyramidal

syndrome, which was analyzed in a sample of 289 children, 199 of whom presented tremors after suffering a TBI.[18] In neuroimaging tests, the lack of consistent findings in CT and MRI may infers that the lesions responsible for tremor are of biochemical nature.[20] In the present case, although there were no changes in the initial CT, the MRI of the head revealed alterations consistent with bilateral gliosis areas in the putamen and in the globus pallidus. Iwadate et al, in a case series of Japanese patients who suffered severe TBI, also observed a predominance of midbrain lesions, even though the patients presented late diffuse brain lesions in CT scans.[24]

Regarding the drug therapy for extrapyramidal syndrome resulting from TBI, papers dealing with the subject describe the use of different pharmacological groups. Some cases were successfully treated with benzodiazepines, β-blockers and anticonvulsants.[20] A study in children with post-TBI tremor showed that propranolol therapy was satisfactory for symptom control.[20] Drake et al also obtained a satisfactory control with the use of benzodiazepines and anticonvulsants, such as phenobarbital, carbamazepine, and phenytoin.[15] In the present case report, haloperidol lead to a good control of the symptoms. The drugs used in the conservative treatment of the tremor belong to different pharmacological groups. Some drugs act on different ion channels present in the axons, reducing the triggering of action potentials. Others act by reducing the activity of the sympathetic autonomic nervous system, and some influence the dopaminergic system, interfering directly in the circuits between the basal ganglia and the cerebral cortex.[25] Another differentiation should also be considered. Haloperidol is an antipsychotic of the butyrophenone group that blocks postsynaptic mesolimbic D1 and D2 dopaminergic receptors.[25] Propranolol, an antihypertensive and non-selective blocker of β1 and β2 receptors, can reduce the activity of the autonomic sympathetic nervous system.[25] It is noteworthy that the mechanism of action of the anticonvulsant phenobarbital is not yet fully known. This drug probably binds to an allosteric regulatory site at the γ-aminobutyric acid $_A$ ($GABA_A$) receptor, extending the opening time of the chloride ion (Cl^-) channels.[25] Similarly, phenytoin acts as an anticonvulsant, and its mechanism of action is the reduction of the triggering of action potentials by blocking voltage-activated sodium (Naþ) channels.[25] Finally, the mechanism of action of carbamazepine is based on sodium channels blockade, which also reduces the triggering of action potentials.[25]

Among the surgical options, deep cerebral stimulation of the thalamus is highlighted, more precisely in the ventro-oralis posterior/ventral intermedius nuclei (Vop/Vim) complex, with satisfactory control of the symptoms.[26] Martínez-Mañas et al achieved a good control in a teenager presenting with a mixed hyperkinetic syndrome manifested by tremor, hemiballism and chorea that were refractory to oral medication.[26] Andrew et al, in a case series of eight patients with extrapyramidal syndrome due to TBI, also obtained a good control of the symptoms through thalamotomy performed through stereotaxis in the Vim complex.[20,27] In this series, some of the patients developed, as a postoperative complication, a worsening of the dysarthria and of the hemiparesis already present before the procedure.[20,27] It is important to emphasize that deep brain stimulation has been a form of non-ablative and reversible treatment when compared with stereotactic lesionectomy. Sitsapesan et al showed the feasibility of post-TBI tremor treatment through deep cerebral stimulation. The work of these authors included eight patients whose brain stimulation involved the posterior Vop and an uncertain zone with a control of the severity of symptoms of $\sim$ 80%.[28]

Regarding the prognosis of the patients with extrapyramidal syndrome resulting from TBI, it was noted that, in half of the cases of a large series, the symptoms disappeared spontaneously over time.[18]

Conclusions

Traumatic brain injury sequelae revealed by extrapyramidal syndrome are often rare. Their pathophysiology is not well understood yet, and it is possibly associated with basal ganglia and/or midbrain lesions. In several cases, conservative treatment with different drugs, such as benzodiazepines, anticonvulsants and β-blockers, was performed. Among the surgical treatments, stereotactic thalamotomy and, more recently, deep brain stimulation, are highlighted. The patient whose case is described in the present study presented a satisfactory control of the tremor with haloperidol. Thus, the optimal therapy for post-TBI extrapyramidal syndrome control is yet to be well defined, thus requiring new studies and broad discussions.

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Ultrasonographic Evaluation of the Optic Nerve Sheath in the Diagnosis of Idiopathic Intracranial Hypertension

Ultrassonografia da bainha do nervo óptico no diagnóstico da hipertensão intracraniana idiopática

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Abstract

Intracranial hypertension (ICH) is a life-threatening condition that can be observed in several diseases. Its clinical presentation is variable, with headache, nausea, vomiting, visual disturbances, papilledema, and alterations in the level of consciousness. The gold standard for the diagnosis of ICH is still the intracranial implantation of invasive devices. Non-invasive techniques, such as ultrasonography of the optic nerve sheath (USONS), have emerged in recent years with promising clinical results. The authors report the case of a patient with progressive headache associated with visual impairment and papilledema, and the eventual diagnosis of idiopathic intracranial hypertension using USONS.

Keywords
- intracranial hypertension
- ultrasonography
- optic nerve sheath

Resumo

A hipertensão intracraniana (HIC) é uma condição clínica potencialmente grave, podendo ser observada na vigência de vários processos patológicos. O quadro clínico pode se manifestar com cefaleia, vômito, alterações do nível de consciência, alterações visuais e papiledema. O padrão-ouro para o diagnóstico da HIC permanece sendo através de medida invasiva, com a instalação de dispositivo intracraniano. Técnicas não invasivas, como a ultrassonografia da bainha do nervo óptico (USBNO), têm surgido nos últimos anos com resultados promissores na prática clínica. Os autores relatam o caso de um paciente jovem com história de cefaleia progressiva associada a alterações visuais e papiledema que teve o diagnóstico de hipertensão intracraniana idiopática auxiliado pela utilização da USBNO.

Palavras-chave
- hipertensão intracraniana
- ultrassonografia
- bainha do nervo óptico

Introduction

Intracranial hypertension (ICH) is a frequent complication in patients with neurological disorders, and it is associated with high morbidity and mortality. As such, its early diagnosis and the institution of adequate therapeutic measures are fundamental for a good prognosis.[1] These patients often require multimodal monitoring of the intracranial pressure (ICP), of the cerebral perfusion pressure (CPP), of the metabolism and

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tissue oxygen consumption, of the electrical brain activity, and of the body temperature, either by invasive or non-invasive devices.[2] Among the aforementioned variables, ICP measurement and its maintenance at levels < 20 mm Hg is, alone, the most important factor for a good neurological outcome.[3]

The gold standard for the measurement and follow-up of ICP still is the use of intracranial devices, specifically with the implantation of catheters. However, the invasive approach of this technique has multiple disadvantages and potential severe complications, such as bleeding and infections; moreover, it requires the presence of a specialized professional—in this case, a neurosurgeon—that is not available in most services. In many cases, even with the availability of a neurosurgeon, there are contraindications to the procedure, such as bleeding disorders.[4]

In recent years, several noninvasive methods have been developed to provide an alternative for the diagnosis of ICH, such as transcranial neuroimaging and Doppler studies.[8] However, although these methods pose less risk of complications, their accuracy remains limited.

Ultrasonography of the optic nerve sheath (USONS) is a promising diagnostic tool that can be used at the bedside. Since the optic nerve is a continuation of the central nervous system, it is encased by cerebrospinal fluid (CSF). Therefore, if the circulation of CSF is not blocked, the increased ICP is transmitted through the subarachnoid space around the optic nerve, within its sheath, especially in the retrobulbar segment.[12]

The present article reports the utility of USONS in the diagnosis of a patient with idiopathic intracranial hypertension (IIH).

Case Report

A male patient, 25 years old, physician, presented to the neurology clinic with a 10-day history of holocranial headache of mild to moderate intensity and of progressive character, reported as a "headache sensation" and associated with visual changes described as scintillating scotomas and turbidity. A general physical examination showed that the patient was obese (body mass index [BMI] = 48 kg/m^2) and presented violet abdominal striae. The ophthalmologic evaluation with retinography evidenced inaccurate limits of the optic disc bilaterally, with poorly delimited margins and exudates, consistent with papilledema (►**Figs. 1** and **2**). There was no acuity or reduction of the visual field. The neurological examination did not show alteration of consciousness, cranial nerve palsy or focal deficit. General laboratory tests revealed only abnormalities in the cholesterol levels. There was no hormonal disturbance. A magnetic resonance imaging (MRI) exam was requested for etiological investigation and showed an empty sella and increased CSF space in the optic nerve sheath, but no other findings. Due to the clinical suspicion of IIH, an USONS was performed, and its result suggested ICH, with a nerve sheath of 0.52 mm on the right side (►**Fig. 3**) and of 0.54 mm on the left side (the normal reference value adopted in our service is up to 0.48 mm).[13] A lumbar puncture was performed with the patient in left lateral decubitus which

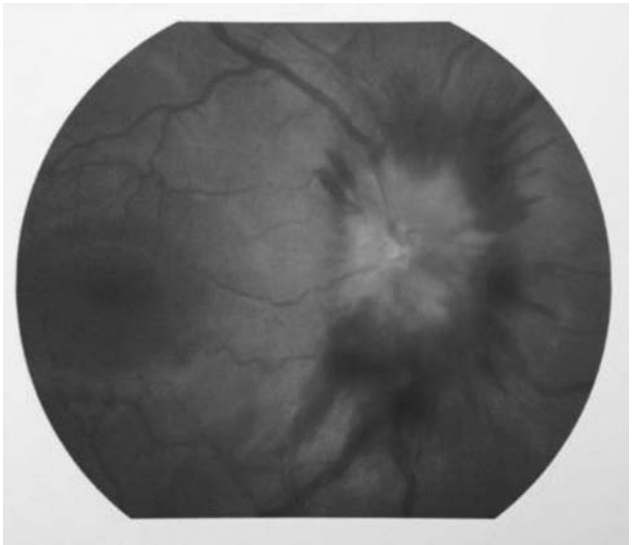

Fig. 1 Retinography—right eye—Optical disc with poorly delimited edges and exudates, consistent with papilledema.

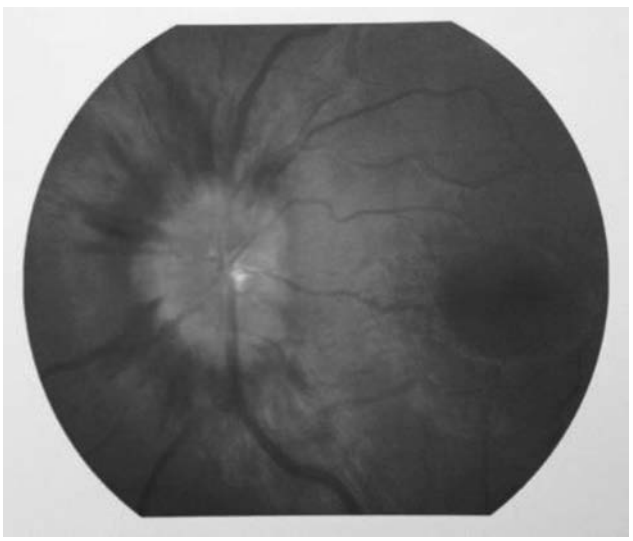

Fig. 2 Retinography—left eye—Optical disc with poorly delimited edges and exudates, consistent with papilledema.

an opening pressure of 36 mm Hg. The analysis of the CSF (cytology, cytometry, total protein, glucose, lactate dehydrogenase, and microbiological tests for bacteria and fungi detection) revealed normal findings, confirming the suggested diagnosis of IIH. Treatment with acetazolamide was started at an initial dose of 750 mg/day (250 mg, 3 times per day) and, as a non-pharmacological measure, the patient was oriented to lose weight through dieting and physical activities; in addition, he should be followed-up by the neurological and ophthalmic services. At a new outpatient visit, 45 days post-treatment the patient reported resolution of the headache and visual changes. An ophthalmologic evaluation with a new retinography showed resolution of the papilledema (►**Figs. 4** and **5**). The patient was under nutritional monitoring, having already lost 8 kg.

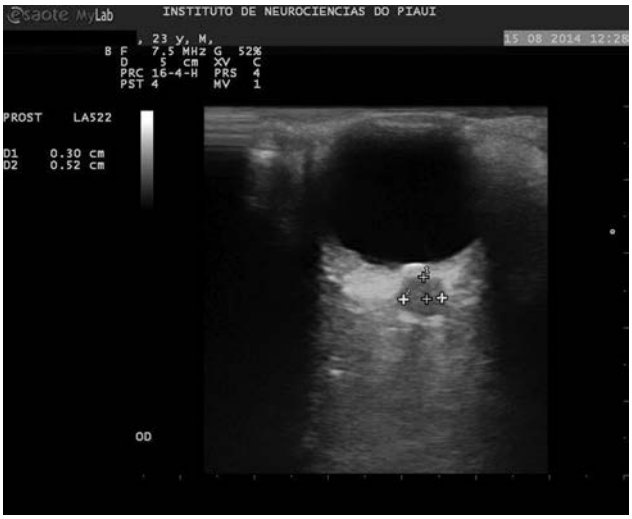

Fig. 3 Ultrasonography of the optic nerve sheath—right eye—Increased diameter of the nerve sheath (diameter = 0.52 mm, reference value adopted at the service = up to 0.48 mm).

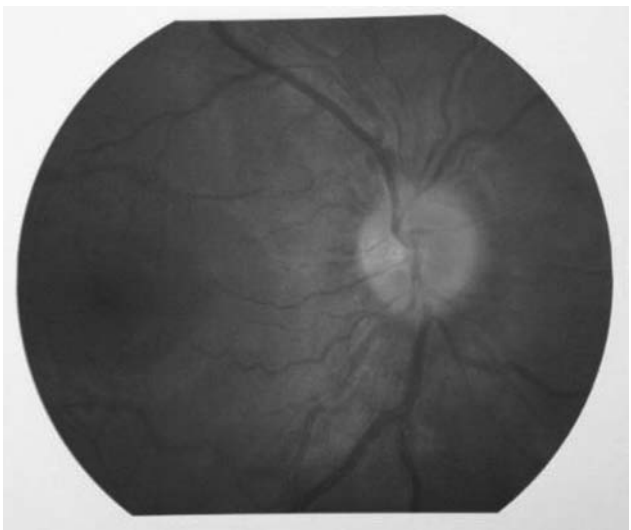

Fig. 4 Retinography after 45 days of treatment—right eye—optic disc with sharp edges and no exudates.

Discussion

Idiopathic intracranial hypertension is a pathology of unknown etiology that affects mainly young, obese women. The fundamental problem in this disease is the chronic elevation of the ICP, and its most important neurological manifestation is papilledema, which can lead to progressive optic atrophy and blindness.[14]

The presentation of a patient with ICH symptoms (headache, visual impairment and papilledema) should be considered a medical emergency, and a neuroimaging examination (preferably MRI) should be performed to investigate the presence of an intracranial expansive lesion. If detectable lesions are absent, the diagnosis of IIH is likely. This pathology is not associated with a specific risk of mortality, but morbidity is observed as a result of incapacitating headache and, specially, visual changes that can progress to blindness.[15,16]

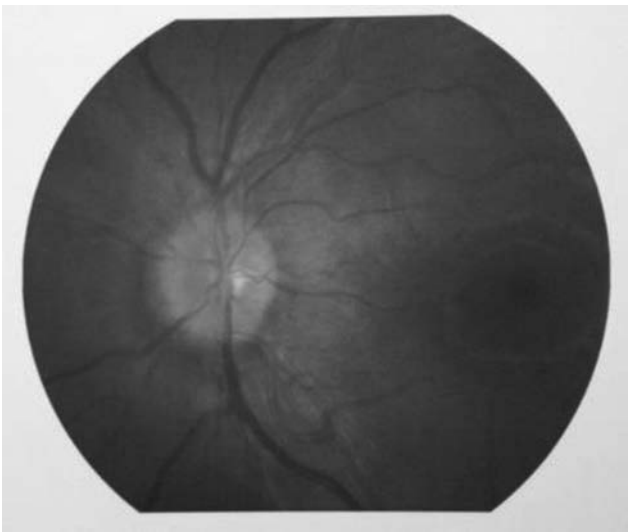

Fig. 5 Retinography after 45 days of treatment—left eye—optic disc with sharp edges and no exudates.

A study by Prunet et al showed that the normal optic nerve sheath diameter (ONSD) ranges from 0.30 to 0.49 mm.[13] Soldatos et al showed that head trauma patients present a proper correlation between ONSD and ICH, demonstrating that values > 0.54 mm correlate with increased ICP values (> 20 mm Hg) with 71% sensitivity and 100% specificity.[17] Roque et al also studied the usefulness of the ONSD measurement, indicating a cutoff diameter of 0.50 mm.[18]

Conclusion

Ultrasonography of the optic nerve sheath is a non-invasive technique, easy to perform at the bedside, with no associated complications, that can be useful in the research and monitoring of patients with clinical suspicion and diagnosis of IIH. It appears as a promising procedure for the evaluation of ICP in various contexts that can coexist with ICH.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Erratum - Effect of Albendazole Treatment in an Experimental Model of Neurocysticercosis-Induced Hydrocephalus

Errata - Efeito do tratamento com albendazol num modelo experimental de hidrocefalia induzida por neurocisticercose

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Arq Bras Neurocir 2019;38:77.

ERRATUM

Rio de Janeiro, February 28, 2019

Dear readers,

In the Article *Effect of Albendazole Treatment in an Experimental Model of Neurocysticercosis-Induced Hydrocephalus* (*DOI: 10.1055/s-0039-1678561*), published online in Arq Bras Neurocir in February 2019, where it reads:

Pedro Tadao Fogaroli

It should read:

Pedro Tadao Hamamoto Filho

ERRATA

Rio de Janeiro, 28 de Fevereiro de 2019

Prezados leitores,

No artigo *Efeito do tratamento com albendazol num modelo experimental de hidrocefalia induzida por neurocisticercose* (*DOI: 10.1055/s-0039-1678561*), publicado online em Arq Bras Neurocir em Fevereiro de 2019, onde se lê:

Pedro Tadao Fogaroli

Lê-se:

Pedro Tadao Hamamoto Filho

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