

ARACHNOIDITIS OSSIFICATIONS IN THE SPINE

ARACNOIDITE OSSIFICANTE NA COLUNA VERTEBRAL

ARACNOIDITIS OSIFICANTE EN LA COLUMNA VERTEBRAL

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ABSTRACT

Introduction: Arachnoiditis ossificans (AO) in the spine is a rare entity characterized by progressive calcification of the arachnoid and dural sac, with consequent neurological involvement. **Objective:** Review the causes, clinical manifestations, and complementary studies for their correct diagnosis. **Method:** Systematic review under PRISMA guidelines, with search in Pubmed, Lilacs, and Embase. Patient demographics (sex and age), history reported as a cause of AO and time elapsed between cause and diagnosis of AO, clinical manifestations, and complementary studies used for diagnosis were collected. **Results:** 38 articles, of which we collected 46 patients (25 women, 21 men), mean age of 52 years. The most frequent cause was previous spine surgery and myelography with fat-soluble contrast. The most frequent symptoms were insufficient muscle strength (74%) and pain (69%). CT was used in 76%. The most frequent location was thoracic (35%). **Conclusions:** Its pathogenesis is unclear; described as the final cause of a chronic inflammatory process in the arachnoid with the consequent bone metaplasia. Diagnosis usually precedes a long period of pain and progressive neurological symptoms. The most sensitive and specific complementary study for the diagnosis is the tomography without contrast, which should be requested in case of clinical suspicion. **Level of Evidence II; Systematic Review.**

Keywords: Arachnoiditis; Spine; Myelography; Pain; Tomography.

RESUMO

Introdução: A aracnoidite ossificante (AO) na coluna vertebral é uma entidade rara caracterizada por calcificação progressiva do saco aracnóideo e dural, com consequente envolvimento neurológico. **Objetivo:** revisar as causas, manifestações clínicas e estudos complementares para o seu correto diagnóstico. **Método:** Revisão sistemática sob as diretrizes do PRISMA, com busca no Pubmed, Lilacs e Embase. Foram coletados dados demográficos dos pacientes (sexo e idade), história relatada como causa de AO e tempo decorrido entre a causa e o diagnóstico de AO, manifestações clínicas e estudos complementares utilizados para o diagnóstico. **Resultados:** 38 artigos, dos quais foram coletados 46 pacientes (25 mulheres, 21 homens), com idade média de 52 anos. A causa mais frequente foi cirurgia prévia da coluna vertebral e mielografia com contraste lipossolúvel. Os sintomas mais frequentes foram comprometimento da força muscular (74%) e dor (69%). A TC foi utilizada em 76%. A localização mais frequente foi torácica (35%). **Conclusões:** Sua patogênese não é clara, é descrita como a causa final de um processo inflamatório crônico na aracnoide com a consequente metaplasia óssea. O diagnóstico é geralmente precedido por um longo período de dor acompanhado por sintomas neurológicos progressivos. O estudo complementar mais sensível e específico para o seu diagnóstico é a tomografia sem contraste, que deve ser solicitada em caso de suspeita clínica. **Nível de Evidência II; Revisão sistemática.**

Descritores: Aracnoidite; Coluna vertebral; Mielografia; Dor; Tomografia.

RESUMEN

Introducción: La aracnoiditis osificante (AO) en la columna vertebral es una entidad rara que se caracteriza por la calcificación progresiva de la aracnoides y el saco dural, con la consecuente afectación neurológica. **Objetivo:** revisar las causas, manifestaciones clínicas y los estudios complementarios para su correcto diagnóstico. **Método:** Revisión sistemática bajo las directrices PRISMA, con búsqueda en Pubmed, Lilacs y Embase. Se recolectaron los datos demográficos de los pacientes (sexo y edad), el antecedente reportado como causa de AO y el tiempo transcurrido entre causa y el diagnóstico de AO, las manifestaciones clínicas y los estudios complementarios utilizados para el diagnóstico. **Resultados:** 38 artículos, de los cuales recolectamos 46 pacientes (25 mujeres, 21 hombres), promedio de edad 52 años. La causa más frecuente fue la cirugía previa de columna vertebral y la mielografía con contraste liposoluble. Los síntomas más frecuentes fueron la alteración de la fuerza muscular (74%) y dolor (69%). Se utilizó TC en un 76%. La ubicación más frecuente fue torácica (35%). **Conclusiones:** Su patogenia no es clara, se describe como causa final de un proceso inflamatorio crónico en la aracnoides con la consecuente metaplasia ósea. El diagnóstico generalmente se precede de un largo periodo de dolor acompañado de síntomas neurológicos progresivos. El estudio complementario más sensible y específico para su diagnóstico es la tomografía sin contraste, que debe ser solicitado ante la sospecha clínica. **Nivel de Evidencia II; Revisión sistemática.**

Descriptores: Aracnoiditis; Columna vertebral; Mielografía; Dolor; Tomografía.



INTRODUCTION

Arachnoiditis osificans (AO) in the vertebral column is a rare entity characterized by progressive calcification of the arachnoid and dural sac (Figure 1), with consequent neurological affectation. It should be differentiated from the frequent finding in autopsies of calcified plaques in the meninges (between 43 and 76%), which are attributed to degenerative processes.¹⁻³

The most frequent location is in the thoracic spine, constituting between 66 and 90% of cases, followed by the lumbar location.³⁻⁶

The exact pathogenesis of the calcification process is still unclear.⁷ It is considered the final stage of adhesive arachnoiditis, secondary to a chronic inflammatory process that triggers bone metaplasia in the arachnoid and the consequent appearance of calcified plaques.^{2,8} Multiple factors have been reported to cause this condition, including spinal trauma, syringomyelia, previous neck surgeries, subarachnoid hemorrhage, anesthetic injections and intrathecal medication, myelography with liposoluble contrasts, infections, vascular malformations or intradural tumors, and even endocrinological causes.^{3,7,9}

The neurological symptoms are a consequence of the direct compression of the calcic formations, mainly in the form of plaques several millimeters thick with a ring or tubular distribution, or may be found surrounding the neural structures, as well as producing chronic irritation and irritation of the roots, and occasionally may be associated with the presence of syringomyelia due to CSF flow alteration.⁷

Patients report long periods of low back pain, lumbocatalgia, or signs of progressive myelopathy that may be related to the calcifications' location and severity.^{1,5}

The article aims to review the causes of AO described in the literature, the clinical manifestations, and the complementary studies for its correct diagnosis. Thus, secondarily, providing spinal surgeons with tools for diagnosing and managing the pathology.

MATERIALS AND METHODS

This systematic review was performed under the PRISMA guidelines.

Selection criteria

Articles from patients with a diagnosis of arachnoiditis ossificans are included.

Exclusion criteria

Excluded were studies whose location was extra vertebral,

asymptomatic patients, who did not report data on the causes, the patient's clinic, or complementary studies performed, and cadaver studies.

Search and selection strategy for studies

The bibliographic databases Pubmed, Lilacs, and Embase were searched. The terms "arachnoiditis ossificans," "ossifying arachnoiditis," and "arachnoiditis ossificans" were used, and the following filters were applied: human, Spanish, English, and Portuguese.

Data extraction

Two authors independently extracted and summarized the data from the included studies. In addition, data were collected on the study design, year of publication, demographic data of the patients (gender and age), the history reported as a cause of AO and the time elapsed between cause and diagnosis of AO, clinical manifestations, and the complementary studies used for diagnosis.

Data analysis

We analyzed the distribution of AO by sex and age, the frequency of reported cases, the median time between that cause and the onset of symptoms, and the use of complementary studies.

RESULTS

The literature search yielded 120 studies (76 from Pubmed, one from Lilacs, and 43 from Embase). Twelve were excluded in duplicate, 52 in the first screening of titles and abstracts, and 18 in the second screening after reading the full text, leaving 38 articles included.¹⁰⁻³⁸ (Image 2) We obtained 49 patients, of whom we excluded three because they were asymptomatic. Of the 46 patients obtained, 25 were women, and 21 were men, averaging 52 years.

The most frequent cause was previous spinal surgery; 9 had been operated on more than once, and in second place was myelography with liposoluble contrast (Table 1). In 17 cases, it was due to more than one cause, and three had ankylosing spondylitis. In 26 cases, the time elapsed between the cause and diagnosis of AO was 18 years on average. Concerning the clinical data, 32 cases presented with pain (13 lumbar, 16 lumbar radiating to the lower limbs, and 3 thoracic pain), 13 did not report pain, and 1 reported no pain.

As for the neurological examination, 34 patients presented alteration of muscle strength (30 weakness, 4 plegia); 26 patients had alteration of sensibility (15 hypoesthesias, 4 dysesthesias, 3 paresthesias and 2 hyperesthesias); 18 patients presented sphincteric alterations; 23 patients with alteration in the osteotendinous reflexes (13 hyperreflexia, 6 areflexia and 4 hyporeflexia) and 13 of them in turn, present pathological reflexes. Fourteen patients manifested with gait disturbances, 12 with spasticity, and 2 with rigidity. (Table 2)

Regarding the complementary studies used for diagnosis

Table 1. Causes of AO.

Cause	Patients n:46
Unrecognized	2
Spinal hemorrhage	2
Myelography with liposoluble contrast	11
Syringomyelia	3
Spinal trauma	6
Previous spinal surgery	25
Intrathecal medication	3
Thoracic Hernia	1
Lumbar puncture	3
Parathyroid adenoma	1
Tuberculosis	1
Subarachnoid hemorrhage	3
Epidural anesthesia	3

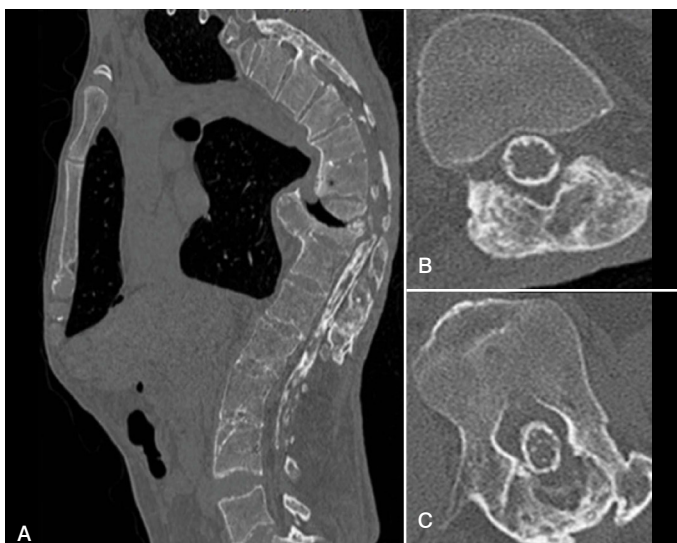


Figure 1. Multislice tomography image of the thoracic and lumbar spine. A- Sagittal section showing intradural calcifications from T2 to S1; B- Axial section showing "circular pattern" at the thoracolumbar level; C- Axial section showing "squat pan pattern" from L3-S1.

Table 2. Clinical manifestations of patients at the time of diagnosis of AO.

Clinical manifestations	Patients n:46
Pain	32 (69%)
Alteration in muscle strength	34 (74%)
Sensitivity alteration	26 (56.5%)
Sphinter Alteration	18 (39%)
OT reflex alteration	23 (50%)
Spasticity	12 (26%)
Rigidity	2 (4%)
Change of gait	14 (34%)

(Table 3), computed tomography (CT) was the most frequently used study (35 patients), followed by MRI (25 patients). However, in 72% of the cases, more than one study was used, and the combination of MRI and CT was the most frequent (22 patients).

The AO location (Table 4) was mainly thoracic, with 16 patients, followed by the lumbosacral location, with 12 patients. Of the patients who had undergone MRI, nine had associated syringomyelia.

DISCUSSION

Although the pathogenesis of AO is unclear, it is described as the final cause of a chronic inflammatory process in the arachnoid with consequent bone metaplasia.³⁹ The causes are varied, but all are associated with epidural or subdural (subarachnoid space) aggressions that consequently induce this process.⁴⁰ In our review, the most frequent causes reported were previous spine surgery and myelography with liposoluble contrast.

The severity and progression of symptoms are varied; diagnosis is usually preceded by a long period of pain accompanied by progressive neurological symptoms. In our review, the median time between the cause and diagnosis was 18 years, where 36% of the articles referred to progressive symptoms. The predominant symptom was an alteration of muscular strength, manifested as weakness in 88% and 12%, with a progressive evolution to paraplegia. According to Maulucci,⁵ the symptoms are more severe when the location is thoracic and less severe when it is lumbar. Although we did not analyze the association between the patient's clinical condition and the location of the AO, we found that 69% of patients with a thoracic column location manifested four or more symptoms and only 28% with a lumbar and lumbosacral location.

The most sensitive and specific complementary study for the diagnosis of AO is the non-contrast CT scan since it allows the evaluation of the bone growth pattern, its extent, and the distribution of neural structures.⁴¹ However, in the complete study of the pathology, MRI is of vital importance to detect the extent of spinal cord compression and the presence of associated pathology such as syringomyelia, arachnoid cysts or tumors; linear or globular lesions hyperintense on T1 and hyper- or hypointense on T2 will be observed, and the administration of gadolinium would be useful to

Table 3. Complementary studies were used.

Studies used	Patients n: 46
TC	6
RNM	1
RNM and TC	22
RNM and RX	1
TC and RX	5
RNM, myelography, and TC	1
Myelography	6
Myelography and TC	1
Myelography and RX	2

CT: computed tomography; MRI: Nuclear magnetic resonance; X-rays.

Table 4. Location of the AO in the spine.

Location	Patients n:46
Cervicothoracic	1 (2%)
Thoracic	16 (35%)
Thoracolumbar	8 (17.5%)
Lumbar	9 (19.5%)
Lumbosacral	12 (26%)

identify areas of fibrosis resulting from chronic inflammation, which could be sites of new calcifications. Therefore, if the initial study is MRI, as is usually the case, AO should be suspected, and a CT scan should be requested.^{3,5,7,9,42} Before the advent of these two studies, myelography was used, which showed areas of blockage but were not specific because the contrast was confused with the calcified plaques, and the diagnosis was made by finding them intra-surgically.³⁸ In our review, myelography was used as a complementary method until 1995.

CONCLUSION

The spinal surgeon should look for this condition when progressive neurological symptoms, mainly associated with pain and altered muscle strength, exist in patients with a related history, such as previous spinal surgery and myelography with liposoluble contrast. A CT scan should always be requested in case of suspicion, either by clinical findings or by suggestive images in MRI.

Given the low frequency of AO, it is necessary to have criteria for diagnostic suspicion and a correct study protocol to be able to later provide the patient with correct therapeutic management and, as far as possible, prevent the causes.

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