



SYMPTOMATIC ARACHNOIDITIS OSSIFICANS ASSOCIATED WITH SYRINGOMYELIA: A CLINICAL CASE AND A BRIEF LITERATURE REVIEW

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Objective. To present a clinical case of arachnoiditis ossificans associated with syringomyelia and a brief literature review with an emphasis on its etiology, pathogenesis and methods of diagnosis and treatment.

Material and Methods. A clinical case of a 68-year-old patient with symptomatic arachnoiditis ossificans is described. The diagnosis was established on the basis of the results of intraoperative biopsy, histological examination of the resected fragment and confirmed in the post-operative period using CT of the spinal cord, multislice CT myelography, etc. Analysis of the course of the pathology raised the following questions: diagnostic criteria and optimal tactics for treating this disease. A brief review of cases of arachnoiditis ossificans described in the literature for the period from 1982 to the present is given.

Results. A review of cases of ossifying arachnoiditis described in the literature showed that today there is no single tactic for diagnosing and treating this disease. In most cases, the diagnosis is established intraoperatively (65 % of analyzed cases). At the preoperative stage, CT provides reliable visualization of ossification. When choosing therapy, it is necessary to be based on the severity of the clinical picture, the degree of ossification of the arachnoid membrane and the presence of concomitant pathology of the affected spinal cord department (such as syringomyelia).

Conclusion. The presence of a growing neurological deficit should be considered an indication for surgical treatment of patients with arachnoiditis ossificans. The goals of surgery should include decompression of neural structures and restoration of normal cerebrospinal fluid circulation.

Key Words: spinal arachnoiditis, arachnopathy ossificans, calcification, syringomyelia, computed tomography.

Please cite this paper as: Kovalenko RA, Osipova NV, Mineev VA, Mitrofanova LB. Symptomatic arachnoiditis ossificans associated with syringomyelia: a clinical case and a brief literature review. *Hir. Pozvonoc.* 2023;20(1):93–101. In Russian.

DOI: <http://dx.doi.org/10.14531/ss2023.1.93-101>.

The first reference to spinal adhesive arachnoiditis is found in the studies of the German neurosurgeon Krause, published in 1907 [1]. From that time, a certain number of papers appeared in the world literature, where the authors presented the cases with this pathology and tried to consider the main causes and patterns of its occurrence, as well as identify the most preferred algorithm for the diagnosis and treatment of the disease in such patients.

Despite the development of diagnostics as well as the study of this pathology, the outcomes of conservative and surgical treatment options remain ambiguous. Therefore, the problem of developing a single algorithm for diagnosis and treatment remains unresolved even now. In addition, the absence of specific clinical manifestations of the disease often leads to a wrong preliminary diagnosis such

as an intramedullary space-occupying lesion.

The objective is to present a clinical case of arachnoiditis ossificans associated with syringomyelia and a brief literature review with an emphasis on its etiology, pathogenesis and methods of diagnosis and treatment.

Case history. Patient L., female, 68 years old, was referred for surgical treatment to the federal center with a diagnosis of intramedullary space-occupying lesion in the thoracic spine. At the time of admission to the hospital, the patient complained of aching pains in the thoracic spine, which increased with physical activity and weakened only in the supine position; burning and creeping sensations along the thoracic spine; and gait impairment. The complaints persisted for a year; conservative treatment had no effect. When collecting anamnesis,

the patient reported a home injury that occurred about five years ago. She fell from a height of 1 meter onto her back with a blow to the thoracic spine.

The neurological status at admission to the hospital revealed foot paresis up to four points and dissociated disorders of the sensitivity of the lower extremities (impaired superficial pain and temperature sensitivity with preserved deep vibration sensation). There was no dysfunction of the pelvic organs.

According to the findings of contrast-enhanced MRI of the thoracic spine, MR signs of spinal cord structure disorders were detected in the form of a diffuse inhomogeneous increase in the MR signal intensity on the T2-WI at the level of the T4–T11 vertebral bodies (Fig. 1a). Syringomyelia was found at the level of T4–T11 vertebrae with a maximum size of 6 × 4 × 13 mm (Fig. 1b). On the T7–

T9 segment, signs of weak pathological heterogeneous accumulation of a contrast agent were revealed. Attention was drawn to the increased size of the spinal cord throughout the pathological signal and the absence of obvious external compression factors. The situation was regarded as a possible diffuse glial space-occupying lesion (most likely, an astrocytoma of low-grade malignance).

It was decided to perform a laminectomy of the T8 vertebra and a spinal cord biopsy in the area of contrast accumulation.

Intraoperatively, at the T8 level, massive ossification of the dura mater was visualized throughout the posterior semiperimeter. After its dissection, attention was drawn to the presence of an adhesive process and the absence of the flow of cerebrospinal fluid. It was decided not to perform a spinal cord biopsy. Dissection and removal of the ossification fragment were performed along with the sampling of the abnormal membranes of the spinal cord for microscopic examination. The defect in the dura mater was sealed with a TachoComb sponge.

In the postoperative period, considering the intraoperative picture of ossification of the dura mater, MSCT of the thoracic and lumbar spine was performed to clarify the area of ossification. The scan showed areas of calcification and ossification of the spinal cord membranes at the level of T4–T11 vertebrae (Fig. 2a).

To evaluate the patency of the cerebrospinal fluid spaces, a multislice CT myelography of the thoracic and lumbosacral spine was performed. According to its findings, there was a difficult passage of contrast along the posterior and lateral surfaces of the spinal cord from the level of T11 with a complete block at T9 (Fig. 2b).

In the analysis of cerebrospinal fluid, an increase in protein to 0.90 g/L (the normal range is 0.15–0.45 g/L) was observed.

To identify possible metabolic disorders that could lead to calcium deposits in noncharacteristic areas, the patient was prescribed additional laboratory assays: the level of total calcium, phosphorus, sodium, magnesium, thyroid-

stimulating and parathyroid hormones, and free thyroxine. All the studied indicators were within the reference values.

Histological examination of the resected fragment (Fig. 3) showed soft fibrous tissue with sinusoidal vessels (in the right), hyalinized tissue (in the center; indicated by the yellow arrow), and normal bone tissue with trabeculae and marrow adipose tissue (indicated by the blue arrow). Deposits of calcium crystals or proliferation of inflammatory cells (white blood cells and neutrophils) were not detected.

In the postoperative period, there was pain attenuation and the absence of an increase in focal neurological symptoms. Considering moderately pronounced neurological disorders, the preservation of the usual level of activity, and the extent of the pathological process, and the possible preservation of existing disorders or their progression as a result of extensive surgery with decompression throughout ossification, it was decided to desist from performing an extended volume of surgery. Due to the treatment and satisfactory condition, the patient was discharged

from the hospital on the 7th day after the surgery.

Discussion

We reviewed 30 cases of arachnoiditis ossificans described in the world literature from 1982 to 2021 and one case of treatment and follow-up of a patient with this pathology from our own experience.

Cumulative findings: arachnoiditis ossificans was slightly more common in women (58 % of cases) than in men (42 % of cases); the earliest age of a patient diagnosed with arachnoiditis ossificans was 13 years old, the latest was 89 years old; the pathology was most common in the age group from 60 to 69 years (32 % of cases); the thoracic spine was more often affected (71 % of cases) than the lumbar spine (29 % of cases), there were no cases of cervical lesions; the presence of a history of spine and spinal cord injuries (domestic or iatrogenic), subarachnoid hemorrhage, neuroinfections and other possible risk factors for arachnoiditis was noted in 81 % of cases; syringomyelia was found



Fig. 1

MRI of the thoracic spine (T2-WI) of patient L, female, 68 years old: **a** – pathologically altered signal from the spinal cord at the level of T4–T11; **b** – formation of syringomyelic cysts (indicated by arrows)

in 48 % of cases. Among the analyzed cases, the decision on surgical treatment of patients was made in 74 % of cases, improvement after surgery and monitoring the patient's condition after a few months were noted in 64 %. It is also significant to note that in most cases, the diagnosis of arachnoiditis ossificans was established intraoperatively (65 % of the analyzed cases) [2, 3–25].

The selection of sources for inclusion in the study was conducted by searching in the following databases: PubMed, Elsevier, and Google Scholar by the keywords “adhesive”, “ossificans”, and “ossifying arachnoiditis”.

A total of 30 cases were selected with a detailed description of the history of the disease, neuroimaging data, as well as follow-up in the postoperative period of at least 3 months. The observations analyzed under this study are given in Table.

Data on the incidence rate of the spinal arachnoiditis and its occurrence are limited. The incidence is estimated to be 1–2 cases per 10,000 people [26]. Specifically, the number of patients who were diagnosed with arachnoiditis ossificans during their lifetime, according to world literature, does not exceed 90. In this regard, this pathology remains an actual problem of spinal surgery.

As provided by the findings obtained in the diagnosis of arachnoiditis ossificans by CT and MRI, Domenicucci et al. [27] proposed a classification of the disease by radiological signs (form and position of calcification or ossification relative to the dural sac): Type I: banana-shaped or semicircular is a semicircular contour that captures a section of the dural sac and deforms it (found mainly in the thoracic spine); Type II: ring-shaped or circular is a circular contour that captures the entire circumference of the dural sac (found in both thoracic and lumbar spine); Type III: honeycomb – capture of the entire contents of the dural sac at the affected level (found only in the lumbar spine) [27].

The pathogenesis of spinal arachnoiditis is based on the infectious or aseptic inflammation of the spinal arachnoid mater. In most cases, arachnoiditis is a consequence of spinal cord injury



Fig. 2

MSCT of the thoracic spine of patient L, female, 68 years old: **a** – foci of ossification of the dura mater; **b** – MSCT myelography

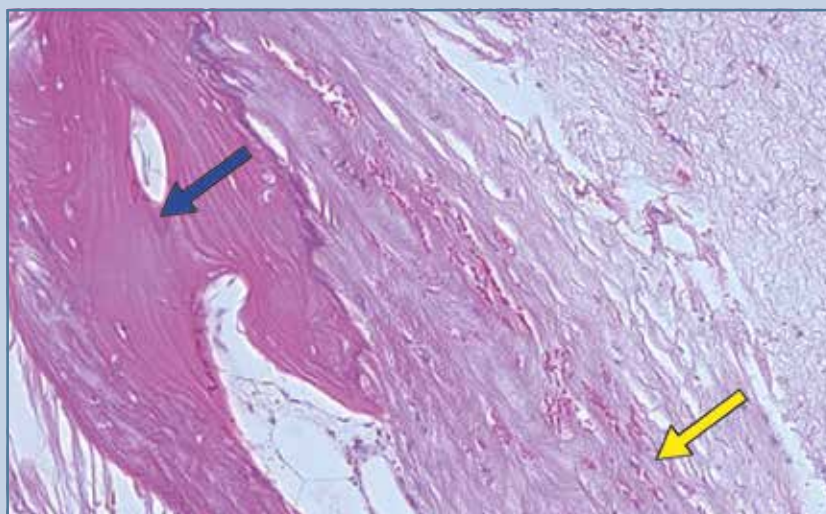


Fig. 3

Surgical specimen: a fragment of ossificate with altered maters of the spinal cord; stained with hematoxylin-eosin, mag. 100

(domestic or iatrogenic), subarachnoid hemorrhage resulting from rupture of arteriovenous malformations of spinal cord vessels, generalized forms of the infectious process (tuberculosis, syphilis, etc.), and neuroinfections [1, 2, 26]. Also, a risk factor for the development of inflammation of the arachnoid mater may be the introduction into the subarachnoid cavity for diagnostic and therapeutic purposes of foreign bodies and contrast agents, mainly oil-based, no longer used nowadays [1]. Additionally, cases of chronic adhesive arachnoiditis have

been revealed in several family members at once who did not have a history of events that could have preceded the development of the disease, after which there were assumptions about its inherited forms [28].

Not every inflammation of the arachnoid mater, however, is followed by the formation of calcifications or ossifications in it. There are many different assumed risk factors resulting in the development of both the first and second conditions, and it is essential to differentiate them correctly.

Calcification of the arachnoid mater of the spinal cord is a frequent phenomenon that, according to autopsies, is found in 76 % of cases [3]. This condition is the outcome of the deposition of crystals (for example, hydroxyapatite) and is often associated with the processes of age-related degenerative mineralization, kidney failure, hyperparathyroidism and hypercalcemia in the anamnesis [4]. It often has no particular clinical significance due to the absence of pronounced symptoms.

At the same time, the term “arachnoiditis ossificans” means a rare condition under which there is a chronic proliferative process resulting in osseous metaplasia of the arachnoid mater of the spinal cord [27, 29].

Histologically, arachnoiditis ossificans is characterized by the presence of osteoblasts with clusters of arachnoid cells. It is considered that mature bone originates from arachnoid cells with the ability to perform multipotent differentiation [5]. Therefore, osseous metaplasia can occur secondary to any process that triggers either chronic inflammation (hemorrhage, infection, surgery, myelography, etc.) or the proliferation of arachnoid cells into osteoblasts [2, 27, 30].

Most cases of arachnoiditis ossificans are detected in the thoracic spine. It is assumed that the highest concentration of arachnoid cells is here, i.e., the appearance of osseous metaplasia mainly in the thoracic region is associated with excessive trabeculation of the arachnoid mater [31]. Additionally, the manifestation of ossification may represent the end point of focal adhesive arachnoiditis [2]. Endocrine factors and genetic predisposition may also be relevant. Nevertheless, a review of the literature sources did not show a connection between arachnopathy ossificans and systemic disorders of calcium metabolism in the body [4].

It is also suggested that both dystrophic calcification and ossification can occur at the areas of fibrosis of the arachnoid mater formed at the areas of preexisting lesions [1].

It is necessary to point out that there is some degree of contradiction in the literature sources regarding the termi-

nology used in the description of arachnoiditis ossificans. Some authors include both pathological conditions in the concept of arachnoiditis ossificans; others separate them.

Indeed, there are no ways to distinguish calcification of the arachnoid mater from its ossification at the diagnostic stage. That why researchers believe that the concept of “arachnoiditis ossificans”, that includes both pathologies, is a more precise term.

However, with the help of histological examination, it is possible to distinguish between these two pathological conditions, since a single occurrence of benign calcification and clinically manifested arachnopathy ossificans will have different consequences and approaches to treatment.

In most patients, pain is the initial and often the only sign in the clinical picture [3–8]. It is marked by constant aching pains in the thoracic and lumbar spine and/or in the legs, which increase during physical activity and resolve at rest. Additionally, some patients complained of hypoaesthesia, paresthesia and gait impairment [2, 8–10, 27]. In some cases, there was a dysfunction of the pelvic organs, mainly the bladder, in the form of enuresis [2–4, 7, 10, 11, 27].

The diagnosis of arachnoiditis ossificans is often established intraoperatively and according to results of a biopsy of resected fragments of the affected area of the arachnoid mater. The most informative techniques for early diagnosis of this pathology are CT, MRI of the affected or all regions of the spine, and CT myelography. The protein content in the cerebrospinal fluid may be noticeably enhanced with impaired outflow of cerebrospinal fluid due to arachnoiditis or may remain within the reference values.

For a differential diagnosis between arachnoiditis ossificans and calcification of the arachnoid mater, additional laboratory assays can be performed, including a blood biochemistry test (determination of electrolytes: sodium, calcium, magnesium, phosphorus). If the electrolytes are within normal range, it is possible to exclude general metabolic disorders resulting in the calcium deposit. It is

required to determine the level of parathyroid hormone in the blood: normal indicators of the concentration of the hormone in the blood exclude the presence of hyperparathyroidism and thyrotoxicosis as the most common causes of calcification; a blood test to determine the concentration of thyroid-stimulating hormone and free triiodothyronine and thyroxine to evaluate the function of the thyroid gland; the determination of vitamin D and calcitonin levels.

Due to the rarity of the occurrence of arachnoiditis ossificans, it was impossible to define a standard option for its treatment at the moment. The choice between conservative and surgical therapy performed individually for each patient in described cases in the literature, depending on the severity of the disease, the degree of ossification of the arachnoid mater, the presence of syringomyelia, and other factors.

Nonetheless, A.A. Zuev et al. [2] proposed the following surgery for patients with arachnoiditis ossificans associated with syringomyelia.

If the length of the ossification does not exceed three segments:

- 1) decompressive laminectomy;
- 2) elimination of spinal cord fixation;
- 3) minimal dissection of the ossified fragment when the clinical picture develops due to local compression of the spinal cord by the ossified arachnoid mater;
- 4) dura mater grafting at fixation levels.

When the ossification length is more than four segments:

- 1) decompressive laminectomy;
- 2) syringo-subarachnoid bypass at the distal part of the cyst into the subarachnoid cavity (in the absence of such a cavity – into the thoracic / abdominal cavity).

The authors believe that the elimination of fixation and dissection of the ossified fragment in the case of extended arachnopathy is inappropriate due to the high risk of an increase in neurological impairment. Additionally, a number of authors advise not to perform extensive surgical intervention to eliminate the long-term effects of surgery in the form of accumulation of a large amount of

Table

Analysis of clinical observations of patients with arachnoiditis ossificans (according to the literature)

Authors	Gender/age, years	Lesion level	Case history	Surgical treatment	Improvement after treatment
Lucchesi et al. [25]	F/64	LS (L4–S1)	Microdiscectomy L4–L5, L5–S1	—	—
Zuev A.A. et al. [2]	F/55	TS	—	T6–T12 laminectomy, elimination of SC fixation, AM resection along the posterior surface of T6–T12, syringo-subarachnoid bypass, grafting of DM	+
	F/60	TS	Removal of neurinoma (T5–T6), drainage of cysts	T4–T8 laminectomy, elimination of SC fixation, AM resection at T6–T12, syringo-subarachnoid bypass, grafting of DM	— (without changes)
	M/66	TS	LOF decompression, grafting of DM	T5–T7 laminectomy, elimination of SC fixation, AM resection at T6–T12, syringo-subarachnoid bypass, grafting of DM	— (without changes)
Revilla et al. [3]	F/68	TS	AVM in PCF, subarachnoid hemorrhage	T2–T5 laminectomy, elimination of SC fixation, AM resection	+
Whittle et al. [4]	F/58	TS	Injury as a result of an accident	T5–T8 laminectomy, elimination of SC fixation, AM resection	+
	F/57	LS	Injury caused by a fall from height, hemorrhage in the SC	L2–L3 laminectomy, elimination of SC fixation, AM resection	— (without changes)
Toribatake et al. [5]	M/65	TS	Meningitis after malaria infection	T4–T9 laminectomy, elimination of SC fixation, AM resection	+
Chan et al. [6]	F/35	TS	Spinal cervicothoracic dermoid cyst, surgical removal	—	—
Singh et al. [7]	F/81	TS	—	T6–T8 laminectomy, elimination of SC fixation, AM resection	+
Wang et al. [8]	F/70	TS	Oil myelography	T5–T8 laminectomy, elimination of SC fixation, AM resection, syringo-subarachnoid bypass	+
Bailey et al. [9]	F/68	TS	Discectomy T2–T3	T2–T9 laminectomy, elimination of SC fixation, AM resection	+
Brunner et al. [10]	M/55	LS	—	L5–S1 laminectomy, durotomy	+

The end of the Table					
Kasai et al. [11]	M/13	LSS	—	Laminectomy at L5–S1, subtotal resection of the arachnoid mater and complete removal of the ossificate	+
Bagley et al. [12]	F/48	TS	Basilar artery aneurysm, C1–C2 laminectomy, the 4th ventricle bypass	T1–T11 laminectomy, elimination of SC fixation, AM resection, grafting of DM	+
Barthelemy et al. [13]	M/43	TS	T5 fracture	—	—
Capron et al. [14]	M/66	TS	Arachnoid cyst at T7 level	T7–L1 decompression, syringo-subarachnoid bypass	+
Faure et al. [15]	M/38	LS	+	—	+
Ibrahim et al. [16]	M/36	TS	Head injury	T4–T7 laminectomy, elimination of SC fixation, AM resection	+
Jaspan et al. [17]	M/42	LS	Myelography, L4–S1 decompression	—	—
	F/41	LS	Myelography, L4–S1 decompression	—	—
	F/50	LS	Myelography, discectomy	—	—
Kahler et al. [18]	M/62	TS	Aneurysm of the left VA, SAH	T2–T11 laminectomy, elimination of SC fixation, AM resection, syringo-subarachnoid bypass	+
Mello et al. [19]	F/49	TS	T10 meningioma, myelography	T10 laminectomy, elimination of SC fixation, AM resection	—
	F/47	TS	Meningitis	T2–T6 laminectomy, elimination of SC fixation, AM partial resection	—
Opalak et al. [20]	M/62	TS, LS	Injury, surgery	T11–L2 laminectomy, elimination of SC fixation, AM resection, grafting DM	+
Papavlasopoulos et al. [21]	M/30	TS	—	Laminectomy at T8–T10, without removal of ossification, cyst drainage	—
Slavin et al. [22]	F/54	TS	—	T9–T11 laminectomy, elimination of SC fixation, AM resection, subarachnoid-subcutaneous bypass	—
Steel et al. [23]	M/89	LS	LS injury, microdiscectomy	—	—
Van Paeschen et al. [24]	F/34	TS	Tuberculous meningitis	Decompression at the level of ossification	—

LS — lumbar spine; TS — thoracic spine; LSS — lumbosacral spine; LOF - large occipital foramen; DM — dura mater; AVM — arteriovenous malformation; PCF — posterior cranial fossa; SC — spinal cord; VA — vertebral artery; SAH — subarachnoid hemorrhage; AM — arachnoid mater.

scar tissue and, as a consequence, deterioration of the patient's condition [1, 2].

In the described case, an isolated decompression laminectomy throughout the lesion is, in our opinion, not advisable. This is connected with the fact that the pathological changes are caused by the ossification of the maters with the development of adhesions and impaired cerebrospinal fluid dynamics, not by external compression factors. Removal of the ossificate with dissection of adhesions does not provide clinical improvement but creates difficulties in the grafting of the resulting defect of the dura

mater and is associated with the risk of progression of neurological impairment.

Conclusion

Considering the low incidence rate of pathology, there are objective challenges in the diagnosis of arachnoiditis ossificans. They are associated with the absence of specific clinical manifestations and similarity with other pathological processes (syringomyelia, space-occupying lesions of the spinal cord, etc.). An indication for surgical treatment of patients with arachnoiditis

ossificans should be regarded as an increasing neurological impairment. The purposes of surgery should include nerve decompression and restoration of normal cerebrospinal fluid flow.

The study had no sponsors.

The authors declare that they have no conflict of interest.

The study was approved by the local ethical committees of the institutions.

All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.

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Received 23.12.2022

Review completed 22.02.2023

Passed for printing 28.02.2023

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