VENTRAL EXTRADURAL SPINAL MENINGEAL CYST CAUSING CORD COMPRESSION

Neurosurgical treatment

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RESUMO - Cistos meníngeos extradurais espinhais são formados tipicamente por estreita cápsula membranosa fibrótica, macroscopicamente semelhante a uma membrana de aracnóide, repleta de líquor e relacionada com uma raiz nervosa ou com a linha média posterior. Eles são extremamente raros em posição anterior e, quando ocorrem, habitualmente causam herniação da medula espinhal pela falha dural ventral. O caso de um homem de 61 anos de idade que iniciou com tetraparesia, espasticidade e hiperreflexia em membros inferiores, e flacidez com hipotrofia nos membros superiores, sem manifestação sensitiva, é apresentado. A investigação com ressonância magnética demonstrou extensa coleção cística extradural ventral à medula de C6 a T11. A lesão foi abordada diretamente via laminectomia com introdução de derivação cistoperitoneal, reduzindo o cisto e tornando o paciente assintomático com um seguimento de 48 meses. Este é o primeiro caso relatado de cisto meníngeo extradural ventral espontâneo causando compressão medular. A derivação cisto-peritoneal se mostrou eficaz no tratamento do caso e deve ser considerada em situações em que a ressecção completa do cisto esteja impossibilitada, ou dificultada pela necessidade de manobras cirúrgicas mais agressivas e arriscadas.

PALAVRAS-CHAVE: cisto meníngeo, cisto aracnóide, lesões extradurais, derivação cisto-peritoneal.

Cisto meníngeo extradural ventral do canal espinhal causando compressão medular: tratamento neurocirúrgico

ABSTRACT - Spinal extradural meningeal cysts are typically formed by a thin fibrotic membranous capsule, macroscopically similar that of an arachnoid membrane, filled by cerebro spinal fluid and related to a nerve root or to the posterior midline. Ventral location is extremely rare and when it occurs they usually cause spinal cord herniation through the ventral dural gap. A 61 year-old man who began with a two years long historyof insidious tetraparesis, spasticity and hyperreflexia in lower extremities, and flaccid atrophy of upper limbs, without sensory manifestations, is presented. Investigation through magnetic resonance imaging demonstrated an extensive spinal ventral extradural cystic collection from C6 to T11. The lesion was approached through a laminectomy and a cyst-peritoneal shunt was introduced. The cyst reduced in size significantly and the patient is asymptomatic over a 48 months follow-up. This is the first reported case of a spontaneous ventral extradural spinal meningeal cyst causing cord compression. Cyst-peritoneal shunt was effective in the treatment of the case and it should be considered in cases in which complete resection of the cyst is made more difficult or risky by the need of more aggressive surgical maneuvers.

KEY WORDS: meningeal cysts, arachnoid cysts, extradural lesions, cyst-peritoneal shunts.

Spinal meningeal cysts are rare lesions. Typically they are formed by a thin fibrotic membranous capsule, macroscopically similar to the arachnoid membrane, filled by cerebrospinal fluid (CSF). A layer of arachnoid cells coating the cyst is not always demonstrated, which explains why the designation of meningeal cyst is preferable to that of arachnoid cyst¹. Ventral position is extremely rare regardless

the cyst is intra or extradurally located^{2,3}. Recently, the 11th case of an anterior intradural spinal meningeal cyst has been reported³. All spontaneous anterior extradural spinal meningeal cysts reported in the literature are related to spinal cord herniation through the ventral dural sleeve, and there is no mention to spontaneous ventral extradural cysts causing posterior cord compression⁴⁻⁹.

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A case of a spontaneous giant anterior meningeal cyst located extradurally is presented (type I), extending from C6 down to T11, and determining severemyelopathy due to posterior cord compression without any sign of herniation. Treatment with cyst-peritoneal shunting resulted in complete regression of the lesion and resolution of the clinical picture.

This article was analyzed by ethic commission of Nossa Senhora das Graças Hospital and its publication was authorized by the patient.

CASE

A 61-year-old man, without history of trauma, presented with a two years long history of insidious tetraparesis associated to back and neck pain for the same period, without sensory complaints. Objectively he showed spasticity and hyperreflexia in his lower extremities and muscle atrophy in both hands. A magnetic resonance image (MRI) revealed a cystic lesion, hypo-intense in T1 and hyper-intense in T2-weighted images, located anterior to the spinal cord exerting significant compression and extending from C6 down to T11 (Fig 1). Further work-up included a myelogram, which revealed a posterior spinal



Fig 1. Sagital MRI demonstrating an anterior cystic lesion, to the spinal cord, hypo-intense in T1 and hyper-intense in T2-weight respectively.

c o rd displacement by a anterior mass filling defect (Fig 2A, 2B). CT myelogram, carried out three hours after contrast injection confirmed the extradural location of the cyst and demonstrated its enhancement, suggesting communication with the subarachnoid space, but without defining accurately its fistulous origin (Fig 2C, 2D).

A C5 - T1 laminectomy was performed. The cervical canal was felt to be very tight and surgical maneuvers to reach its anterior segment were considered risky. Therefore, after adequate exploration of dural root sleeves bilaterally, the dura was opened postero laterally, to allow transdural access to the cyst, through ultrasound guidance.

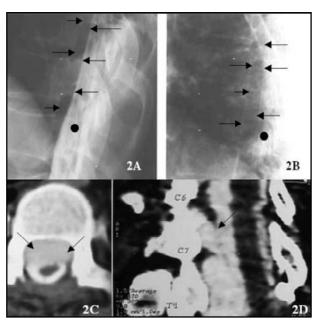


Fig 2. 2A and 2B: Myelogram demonstrating the thecal sac (bla-ck point), and the cyst silhouette (filling defect) seen as a negative image localized behind the vertebral bodies (arrows). 2C: CT-Myelogram done 3 hours after myelogram showing the cyst in an anterior extradural position (arrows). 2D: Reconstruction by CT-Myelogram scan showing the cranial limit of the cyst in C6-C7 and the dural limit between cyst and subarachnoid spaces (arrow), but not defining the exact communication point.

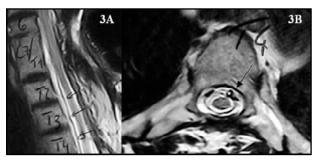


Fig 3. 3A: Sagital T2-weigh MRI demonstrating collapse of the cyst after shunting to the peritoneal cavity (arrows). 3B: Axial image through T4 showing section of drainage catheter (arrow).

The cyst was punctured transdurally and partially emptied, with the total removal of approximately 20 ml of crystal clear liquid resembling CSF. After 20 minutes of observation the cyst refilled. The insertion of a cyst peritoneal drain was then considered then the best surgical option, which proved to be effective in decompressing the spinal cord (Fig 3) and providing total subsidence of signs and symptoms in a 4-year follow-up, as opposed to a more radical and extensive procedure in search for a fistulous communication.

DISCUSSION

Extradural meningeal cysts are typically located posteriorly to the spinal cord and usually originate from a point close to a dural root exit or in the posterior midline¹⁰. Pathophysiological mechanism of the lesion is not very well understood. It could represent the expansion and proliferation of arachnoid granulations through a low resistance dural region^{1,10}, or the occurrence of a congenital or traumatic arachnoid diverticulum, which would expand due to progressive CSF entrance through an osmotic10-13 or an unidirectional valve mechanism11-13. App ropriate characterization of such entity is confusing and redundant in the literature. Nabors and cols. 1 subdivided the meningeal cysts in three groups: I - extradural cysts without associated nervous fibers, II - extradural cysts with associated nervous fibers, and III - intradural cysts. Commonly these lesions are located postero-laterally to the spinal cord, and they can extend along several spinal levels1. Analyzing the various cases of spinal cord ventral hemiation, Kumar and cols.9 suggested a modification in the classification by Nabors and cols.¹, of spinal meningeal cysts to include this mechanism of spinal cord herniation in group II-B9.

The seldom mentioned anterior location of a spinal meningeal extradural cyst may reinforce the congenital or traumatic hypothesis, considering that the anterior dural aspect represents an area of major tissue resistance^{1,10}. Besides, all cases of spontaneous ventral extradural spinal meningeal cyst are related to spinal cord herniation⁴⁻⁹, due to negative pressure exerted by the CSF dynamics inside the cyst over the spinal cord through the ventral dural flaw^{4,8}.

This is the first reported case of a spontaneous ventral extradural spinal meningeal cyst exerting a compressive mass effect on the spinal cord, probably representing a unidirectional valve mechanism through a smaller dural defect. Recently, cyst volume enlargement has been demonstrated by cinematic magnetic resonance studies, associated to higher pressures in the subarachnoid space by Valsalva maneuver¹³.

Among extradural posterior cysts, thoracic location is the most common (67%), followed by lumbosacral (20%), thoracolumbar (9%) and cervical regions (4%). Symptoms are related to cyst compressive effect, flaccid or spastic paraparesis being the most common presentation (70%), which may be accompanied by lumbar or dorsal pain, hyperesthesia, and radiculopathy, usually with a sensory level¹². The patient in the present case had no sensory symptoms, different from most cases reported in the literature, probably because the unusual anterior location of the cyst was mainly related to the motor function portion of the spinal cord.

Myelogram demonstrates findings compatible with an expanding extradural process, which a filling defect¹⁴. In such cases, a late CT-myelogram scan can demonstrate contrast inside the lesion, confirming its communication with the subarachnoid space^{14,15}. MRI is the standard method of choice for diagnosis, defining the location and extension of the cystic lesion, in addition to supply prognostic information about the condition of the spinal cord by the eventual presence of atrophy or signal changes caused by the mass effect^{15,16}. CT-myelogram can aid in determining the communication between the extradural cyst and the subarachnoid space¹⁵. Neither imaging studies nor surgical exploration in the present case were able to demonstrate the origin of the communication, inferred only by refilling of the cyst after its partial emptying and by contrast enhancement.

The proposed ideal surgical treatment to spinal meningeal cysts is cyst resection and closing of dural flaw, in order to avoid the recurrence of fístula^{1,3,10,17,18}. Because the commonest location of these lesions is dorsolateral, a laminectomy usually facilitates cyst dissection in relation to the dura mater. Wide fenestration and shunting of the cyst to the peritoneum, pleural cavity or right atrium are also treatment alternatives^{17,19,20}.

Extension and anterior location of the cyst protruding the cord posteriorly, in the present case, p revented a full exploration, due to risks of damage to spinal cord secondary to excessive manipulation and retraction. It was also felt that shunting the cyst to the subarachnoid space would not solve the problem, considering that communication between both spaces would persist.

The option was shunting the cyst to the peritoneal cavity without interference of a valvular system. The catheter was introduced inside the cyst

through an intradural approach due to difficulties of mobilizing the thecal sac at the cervical level. After its implantation, the system appropriately drained the cyst to the peritoneal cavity with significant reduction of its mass effect on the spinal cord (Fig 3). Total remission of the clinical picture could be demonstrated at a 4 year follow up.

Because all spontaneous ventral extradural spinal meningeal cysts reported in the literature are related to spinal cord herniation through the ventral dural sleeve, surgical treatment usually consisted of excision of the arachnoid cyst whenever it is possible, section of the dentate ligament, release of the adhesions, detachment of the spinal cord from the hernial orifice, and finally suture of the dural tear or placement of a dural patch⁴⁻⁹.

The only case reported in literature related to a ventral spinal cyst treated by a cystoperitoneal shunt is in a 52-year-old man with a previous severe brachial plexus injury. Although a complete resolution of the cyst could be demonstrated, there was no change from his preoperative clinical condition²⁰.

This is the only report of a spontaneous ventral meningeal spinal cyst causing cord compression, adequately treated by shunting to the peritoneal cavity, with an excellent result.

Shunting extradural spinal cysts to peritoneal cavity is an effective treatment. It should be considered as the first choice in situations where location or extension of the lesion prevents its complete resection.

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