Spinal intradural arachnoid cysts located anterior to the cervical spinal cord

Report of two cases and review of the literature

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The authors describe two rare occurrences of radiographically, surgically, and pathologically confirmed spinal intradural arachnoid cysts (not associated with additional pathological entities) that were located anterior to the cervical spinal cord. These lesions have been reported previously in only eight patients. The patients described in this report were young adults who presented with progressive spastic tetraparesis shortly after sustaining mild cervical trauma and in whom no neurological deficit or bone fracture was demonstrated. The presence of an intradural arachnoid cyst was detected on postcontrast computerized tomography (CT) myelography and on magnetic resonance imaging; both diagnostic tools correctly characterized the cystic nature of the lesion. Plain radiography, plain tomography, and contrast-enhanced CT scans were not diagnostic. In both cases a laminectomy was performed, and the wall of the cyst was excised and fenestrated with subarachnoid space. Postoperatively, the patients made complete neurological recoveries.

Based on a review of the literature, arachnoid cysts of the spinal canal may be classified as either extra- or intradural. Intradural arachnoid cysts usually arise posterior to the spinal cord in the thoracic spine region; however, these cysts very rarely develop in the cervical region. The pathogenesis of arachnoid cysts is unclear, although congenital, traumatic and inflammatory causes have been postulated. The authors believe that the formation of an arachnoid cyst cannot be explained by simply one mechanism because, in some reported cases, there has been accidental or iatrogenic trauma in association with congenital lesions. They also note that an intradural arachnoid cyst located anterior to the cervical spinal cord is an extremely rare disorder that may cause progressive myelopathy; however, the postoperative prognosis is good.

KEY WORDS • arachnoid cyst • cervical spine • spinal cord

Cysts of the spinal canal represent an uncommon cause of spinal cord compression and most are arachnoid or diverticula. These cysts have been classified as either extra- or intradural. Intradural arachnoid cysts usually arise posterior to the spinal cord in the thoracic spine region and those located anterior to the spinal cord in the cervical spine region are such rare entities that only eight cases have been reported in the literature. We report an additional two patients in whom an intradural arachnoid cyst (not associated with additional disease) located anterior to the cervical spinal cord was demonstrated. Both patients made a good recovery postoperatively. We also review the literature and report the clinical, neuroradiological, and surgical techniques as well as the results associated with intradural arachnoid cysts located anterior to the cervical spinal cord.

Case Reports

Case 1

Presentation. This 18-year-old man was admitted to our hospital with a 15-day history of progressive weakness in both legs. He had sustained minimal cervical trauma during a basketball game 2 months earlier and had since then been suffering pain between his shoulders and at the back of the neck. Although his pain was slightly reduced when he lay down, it was continuous during the day. Despite the analgesic and antiinflammatory therapy, his symptoms had continued to worsen.

Examination. Neurological examination revealed a mild weakness in extension of both forearms, without involvement of the intrinsic hand muscles, and also a spastic-type paraparesis, more prominent on the left side than the right. Whereas his deep tendon reflexes were decreased in both upper limbs, except for an absent triceps jerk, the deep tendon reflexes in both lower limbs were brisk. A Babinski’s sign and Achilles tendon clonus were also present. Responses to light touch, pin prick, and temperature sense were decreased below the C-6 dermatome. Both joint position and vibration senses were intact. There was no bladder or bowel dysfunction. Results of the remainder of the general physical and neurological examinations were normal.

Plain x-ray films of the cervical spine, computerized tomography (CT) scans, and contrast-enhanced CT scans
of this region demonstrated unremarkable findings. Myelography revealed total block of the subarachnoid space at C6–7 while the patient was in the prone and supine positions (Fig. 1 upper). Postcontrast CT myelography scanning revealed a cystic lesion located anterior to and left of the spinal cord and demonstrated dorsal displacement of the spinal cord at C6–7. Contrast material was not seen to enter the cyst (Fig. 1 lower).

Operation. After induction of general anesthesia, the patient underwent surgery while in a sitting position. A two-level C6–7 laminectomy was performed. The dura and arachnoid membrane were opened in the midline, and the spinal cord was seen to be tense, slightly rotated to the right, and displaced posteriorly. After turning the spinal cord gently by lifting the dentate ligament, an intradural multilobulated arachnoid cyst was revealed that contained arachnoid trabeculations. The fluid that filled the cyst resembled cerebrospinal fluid (CSF). The cyst membranes were partially resected to avoid spinal cord injury and enable the cyst and normal subarachnoid space to communicate freely. Frozen and permanent slides sent for histopathological examination demonstrated an arachnoid cyst.

Postoperative Course. The patient’s postoperative course was uneventful. At discharge, slight spastic paraparesis and a gradual improvement of the sensory disturbances were evident. One month postoperatively his gait was normal. At neurological examination 18 months postoperatively, complete neurological recovery was shown.

Case 2

Presentation. This patient was a 15-year-old boy. He had fallen off a bicycle 15 days prior to presentation. Initially he experienced neck and back pain, and the pain in the back of his neck had gradually increased; it was continuous during the day time and was not altered by musculoskeletal repositioning. Increasing weakness was also apparent in all four extremities, especially on the left side, for the previous 5 days, and he complained of numbness in all extremities. No bladder or bowel dysfunction was reported.

Examination. On neurological examination, he was found to have tetraparesis prominent proximally, especially on the left side. Muscle tone was increased in all extremities without fasciculations or atrophy. Responses to light touch, pin, and temperature sense were decreased below the T-5 dermatome; however, hyperalgesia was observed between the C-3 and T-5 dermatomes. His joint position and vibration senses were normal. His deep tendon reflexes were brisk, and his plantar reflexes were extensor bilaterally. Achilles tendon clonus was present bilaterally. The results of the other neurological and general physical tests were within normal limits.

Plain x-ray films of the cervical spine were unremarkable. Magnetic resonance (MR) imaging revealed an intradural extramedullary cystic lesion located anteriorly to the cervical spinal cord and also demonstrated dorsal displacement and excessive deformation of the spinal cord at C2–3 (Fig. 2 left). The cyst had a well-defined border and did not enhance after contrast administration.

Operation. A left cervical C-3 and C-2 hemilaminectomy was performed with the patient in a sitting position. The dura mater was opened on the left side, and the spinal cord was observed to be rotated slightly to the right and clearly displaced posteriorly. The cyst caused the arachnoid to bulge on the left side of the spinal cord, compressing it anteriorly. Incision of this arachnoid allowed CSF to flow out and the spinal cord to slacken. Using microsurgical techniques, we removed as much of the arachnoid trabeculations and septations that formed the cyst as was feasible, and we fenestrated the cyst with subarachnoid space. We performed duraplasty with the autologous fascia at the end of the operation.

Postoperative Course. The postoperative course was uneventful. Within 2 days postoperatively, we noted an obvious improvement in the patient’s tetraparesis, and he could walk with a cane. Histopathological examination
Anterior cervical arachnoid cysts

![Image]

**TABLE 1**

<table>
<thead>
<tr>
<th>Type</th>
<th>Classification of spinal meningeal cysts*</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>extradural meningeal cysts w/o spinal nerve root fibers</td>
</tr>
<tr>
<td>IA</td>
<td>extradural meningeal cyst (extradural arachnoid cyst)</td>
</tr>
<tr>
<td>IB</td>
<td>sacral meningocele (occult sacral meningocele)</td>
</tr>
<tr>
<td>II</td>
<td>extradural meningeal cysts w/ spinal nerve root fibers</td>
</tr>
<tr>
<td>(Tarlov’s perineurial cyst, spinal nerve root diverticulum)</td>
<td></td>
</tr>
<tr>
<td>III</td>
<td>spinal extradural meningeal cysts (intradural arachnoid cyst)</td>
</tr>
</tbody>
</table>

* The classification is derived from that proposed by Nabors, et al.

Discussion

Terminology and Review of the Literature

Spinal arachnoid cysts have been termed by various authors as “arachnoid diverticula,” “leptomeningeal cysts,” “localized adhesive arachnoiditis,” and “serous spinal meningitis” according to different pathogenetic conceptions. However, Nabors, et al. have called these entities “cysts” for the sake of simplicity and to avoid further confusion in the nomenclature (Table 1). The patient in our cases presented with Type III spinal intradural meningeal cysts (intradural arachnoid cysts) according to this classification.

Intradural arachnoid cysts located anteriorly in the cervical region are extremely rare. In our literature research of the Medline databases comprising reports published between 1966 and February 1999, we retrieved only eight cases in which the intradural arachnoid cyst was located anterior to the cervical spinal cord (Table 2). Alvisi, et al. reported nine patients with intradural arachnoid cysts in the cervical spine region, but all these cysts were located posteriorly or posterolaterally to the spinal cord. Also, intradural arachnoid cysts resulting from spinal cord herniation have been noted in other reports.

Origin of Intradural Arachnoid Cysts

The cause of these cysts remains obscure; some such cysts are ascribed anecdotally to previous trauma or arachnoiditis, whereas the majority are idiopathic and assumed by many authors to be congenital. Aarabi and colleagues have reported two proven cases of intradural arachnoid cysts over two generations in a family with five paraparetic members. The mode of inheritance in this particular family was probably mendelian dominant. It has also been postulated that intradural cysts are a result of a widening of the septum posticum. In the cervical and thoracic spine this septum divides the dorsal subarachnoid space in the midline. However, this hypothesis does not explain cysts that arise on the anterior portion of the spinal cord, such as in our and other cases; it also fails to account for cysts in the lumbar spine region. In addition, Agnoli, et al. have postulated that intradural arachnoid cysts result from a pathological distribution of the arachnoid trabeculae and that this pathological arrangement leads to a diverticulum. The trabecular cells degenerate, and increased pressure builds up within the cyst. The increased pressure has an oncotic effect, and because of the resulting transudation the cyst fills with fluid. At surgery, there were arachnoid trabeculations and septations in our cases. We also established that the cysts were not communicating with subarachnoid spaces in our patients. These findings support the hypothesis of Agnoli, et al. Other possible causes include inflammatory meningitis due to viruses, spirochetes, or bacteria and chemical meningitis secondary to subarachnoid hemorrhage, contrast media, spinal anesthetic agents, and spinal operations. These result in scar formation, which separates the subarachnoid space into diverticula or cysts.

Trauma can be regarded as an accompanying factor. There may not be a primarily closed cyst, and this can be transformed into a cyst gradually via a valvular mechanism. In these cases, trauma can trigger the onset of symptoms through a sudden movement, with a brief increase of CSF pressure from a clinically latent intradural arachnoid cyst. Spiegelmann, et al. have reported that the histological examination of an intradural arachnoid cyst revealed hemosiderin-containing macrophages trapped in the cyst wall; the cyst was therefore considered a delayed consequence of a previous craniospinal injury. In four patients who had suffered mild cervical trauma, however, the time elapsed after the onset of trauma was different in each case (time range 15 days–18 years). In addition, two of three patients with myelomeningocele underwent operations in the craniocervical region for Chiari II malformations. We believe that the formation of these intradural cysts could not be explained by just one mechanism because in these reported cases there had been accidental or iatrogenic trau-
Congenital asymptomatic intradural arachnoid cysts may become symptomatic after traumatic aggravation and extension. Diagnostic Studies

Intradural arachnoid cysts are difficult to diagnose. Plain x-ray films of these spinal lesions are usually not diagnostically helpful. When the cysts occur in the thoracic spine region, radiographic studies can sometimes reveal a kyphosis with signs of long-standing pressure in the form of scalloping of the posterior surfaces of the vertebral bodies, thinning of the pedicles, and widening of the canal. Often the myelographic evidence of an intradural arachnoid cyst is an incidental finding. According to Chan, et al., the most important preoperative diagnostic procedure is myelography performed with the patient in both supine and prone positions. In our first patient, myelography via lumbar puncture revealed a block of contrast material at the C6–7 level with no filling in the cyst, although the patient's position was changed during imaging. Although the radiological appearance suggested an intradural cystic lesion or extramedullary lesion, it was not specific for these. However, postcontrast CT myelography revealed an obvious cystic lesion located anterior to and left of the spinal cord. The cyst was not communicating with subarachnoid space. DiSclafani and Canale have demonstrated a contrast medium–filled posterior mass that compressed the spinal cord by using postcontrast CT myelography. The presence of spinal intradural arachnoid cysts, which may have not been visualized with older techniques, has been demonstrated with modern imaging studies such as MR imaging. However, T2-weighted MR imaging demonstrates a heterogeneous signal intensity, depending on the flow effect in the cyst fluid, which may be the only evidence to indicate the presence of an abnormal fluid collection. In these cases, it has been reported that cine MR imaging can demonstrate abnormal fluid flow and spinal cord compression caused by a spinal intradural arachnoid cyst and also that endoscopy can be a useful tool to diagnose or rule out a cyst.

Patient Management

Surgical treatment is generally indicated for patients with symptomatic spinal intradural arachnoid cysts. Surgical treatment options include excision, fenestration, and placement of a shunt. There is general agreement in the literature that the successful treatment of a spinal arachnoid cyst requires total excision when possible. However, in cases in which the cyst cannot be resected completely because of dense fibrous adhesions to the spinal cord or nerve roots and/or placement anterior to the spinal cord, partial resection and fenestration should be performed as widely as possible. Intradural arachnoid cysts have been reported to recur after incomplete excision. An arachnoid cyst located anterior to the cervical spinal cord has been completely resected in only one of the eight patients in the reported cases. The combined treatment of partial resection, fluid drainage, and fenestration in our two patients produced an excellent return of neurological function in

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**TABLE 2**

**Summary cases reported in the literature on intradural arachnoid cysts located anterior to the cervical spinal cord**

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Trauma (time since onset)</th>
<th>Previous Op/Additional Disease</th>
<th>Symptoms</th>
<th>Level</th>
<th>Surgery</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Palmer, 1974</td>
<td>19, F none</td>
<td>none</td>
<td>spastic tetraparesis</td>
<td>C1–3</td>
<td>laminectomy, drainage, complete resection</td>
<td>improved</td>
<td></td>
</tr>
<tr>
<td>Herskowitz, et al., 1978</td>
<td>3, M none</td>
<td>respiratory infection</td>
<td>tetraparesis</td>
<td>C2–4</td>
<td>laminectomy, repeated fine needle aspiration (3 ops)</td>
<td>death</td>
<td></td>
</tr>
<tr>
<td>Chan, et al., 1985</td>
<td>28, F none</td>
<td>none</td>
<td>tetraparesis</td>
<td>C6–7</td>
<td>laminectomy, drainage, subtotal resection</td>
<td>improved</td>
<td></td>
</tr>
<tr>
<td>Rabb, et al., 1992</td>
<td>37, M cervical soft tissue but no bone fracture (18 yrs)</td>
<td>respiratory infection</td>
<td>tetraparesis</td>
<td>C6–7</td>
<td>laminectomy, cystoperitoneal shunt</td>
<td>improved</td>
<td></td>
</tr>
<tr>
<td>Chen &amp; Chen, 1996</td>
<td>2, F none</td>
<td>myelomeningocele skeletal traction for C-2 fracture (9 yrs)</td>
<td>tetraparesis</td>
<td>C3–5</td>
<td>fenestration, cystopleural shunt</td>
<td>improved</td>
<td></td>
</tr>
<tr>
<td>Jean, et al., 1998</td>
<td>14, F none</td>
<td>myelomeningocele, Chiari II malformation, multiple ventricular shunting, cranioventricular decompression</td>
<td>chronic headaches, vertigo</td>
<td>cervicomedullary junction-C6, C5–6</td>
<td>cystoperitoneal shunt</td>
<td>improved</td>
<td></td>
</tr>
<tr>
<td>present study</td>
<td>18, M cervical soft tissue (2 mos)</td>
<td>myelomeningocele, Chiari II malformation, multiple ventricular shunting, cranioventricular decompression</td>
<td>neck pain, dizziness, tetraplegia, facial muscle weakness</td>
<td>medulla-C5</td>
<td>cyst fenestration w/ subcutaneous reservoir</td>
<td>stabilized</td>
<td></td>
</tr>
<tr>
<td>15, M cervical soft tissue (15 days)</td>
<td>none</td>
<td>spastic tetraparesis</td>
<td>C6–7</td>
<td>laminectomy, partial resection, fenestration</td>
<td>improved</td>
<td></td>
<td></td>
</tr>
<tr>
<td>14, M cervical soft tissue (9 yrs)</td>
<td>none</td>
<td>spastic tetraparesis</td>
<td>C2–3</td>
<td>hemilaminectomy, partial resection, fenestration</td>
<td>improved</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Anterior cervical arachnoid cysts

each case. In the literature, there has only been one death attributed to misdiagnosis and infection. In conclusion, modern imaging techniques make the diagnosis of an arachnoid cyst easier. An intradural arachnoid cyst located anterior to the cervical spinal cord is an extremely rare disorder that may cause progressive myelopathy; however, the postoperative prognosis is good.

References

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