Intrasacral Extradural Arachnoid Cysts
—Three Case Reports—

Mustafa Kemal HAMAMCIIOGLU, Tufan HICDONMEZ, Cumhur KILINCER, and Sebahattin COBANOGLU

Department of Neurosurgery, Trakya University Faculty of Medicine, Edirne, Turkey

Abstract
Three patients presented with rare intrasacral extradural arachnoid cysts manifesting as sensory deficiencies and pain in the lower extremities. Magnetic resonance imaging with various sequences identified the cysts. Two patients underwent surgery via laminectomy of the sacrum for cyst exploration and disconnection of the cyst with the dural theca. Postoperative outcome was favorable in these two patients. Intrasacral extradural arachnoid cyst should be considered in the differential diagnosis of low back pain.

Key words: arachnoid cyst, sacral meningocele, low back pain, magnetic resonance imaging

Introduction
Intrasacral extradural arachnoid cysts were first described as occult intrasacral meningocele based on the myelographic appearance in 1932.2,3,6,11,13,14) The first surgical intervention was performed in 1944.4,5,11,12,14) The description and pathogenesis of intrasacral extradural arachnoid cysts are still controversial, but the origin may be congenital.2,3,5,6,11,13) However, the term meningocele is used to describe herniation of the meninx from a spina bifida or cranium bifidum defect, but the term intrasacral meningocele does not describe a true meningocele.2,3,8,11,14) Some 50 cases have been reported in the literature.1,4,17) Intrasacral extradural arachnoid cysts usually manifest in the third or fourth decade of life as low back pain with or without radiculopathy.6) Perineal pain and sphincteric dysfunction may be present.6,11) Surgery is curative, but there is no consensus on the appropriate surgical treatment method.3–5,10–12,14) We report our clinical experience with three cases of intrasacral extradural arachnoid cyst.

Case Reports

Case 1: A 42-year-old man presented with walking difficulty and pain in the left lower limb. Computed tomography (CT) revealed L4-5 disc herniation. Neurological examination revealed abolished Achilles reflex. Magnetic resonance (MR) imaging revealed an intrasacral extradural meningeal cyst and disc herniations at the L4-5 and L5-S1 levels (Fig. 1). The patient underwent surgical exploration via L5-S3 laminectomies. There was an extradural cerebrospinal fluid-filled cyst extending from L5 to S3 displacing the dural sac anteriorly. The cyst was aspirated and its walls were excised. Its connection to the dural sac was found and primarily repaired. His symptoms resolved just after the operation and did not recur during 9 years of follow up.

Case 2: A 38-year-old man was examined because of complaints of gluteal pain and numbness in the legs for 2 years. Pain increased while standing up and during exercise. Neurological examination and electromyography found no abnormalities. CT and MR imaging revealed an intrasacral extradural meningeal cyst (7 × 8 × 8 cm) (Fig. 2). The patient refused surgical intervention, and although he had intermittent complaints, follow-up examinations were within the normal limits.

Case 3: A 34-year-old woman presented with complaints of low back pain and pain in the left leg, which started 6 years ago after trauma. Neurological examination and CT detected no abnormalities. MR imaging demonstrated an intrasacral extradural meningeal cyst (4 × 1.3 cm) (Fig. 3). Exploration and excision of the extradurally located cyst via S1-S3 laminectomies were carried out and its connection with the dural sac was repaired. She had no complaints in the postoperative period. She remained symptom-free during the 10-year follow-
Fig. 1 Case 1. Sagittal T2-weighted (A) and axial T1-weighted (B) magnetic resonance images showing a large intrasacral cyst. The cyst shows significant widening of the spinal canal and erosion of the sacrum.

Fig. 2 Case 2. Computed tomography scan (A) and T1-weighted non-enhanced axial and sagittal magnetic resonance (MR) images (B, C) showing an intrasacral extradural meningeal cyst (7 × 8 × 8 cm). MR myelogram (D) showing details of the cyst.

Fig. 3 Case 3. Sagittal T1-weighted non-enhanced magnetic resonance image showing an intrasacral extradural meningeal cyst (4 × 1.3 cm).

Discussion

Lumbosacral cysts are classified according to their location, content, and association with dysraphism.6,9,11,14,17) The most common cysts are associated with dysraphic lesions such as meningoceles and meningomyeloceles. Sacral cysts can be classified into three groups: anterior sacral meningocele, intrasacral meningocele, and perineural cysts.6) Another classification of spinal meningeal cysts (Table 1)12) indicates that all three of our cases should be considered as type Ib cysts.

The pathogenesis of intraspinal extradural arachnoid cysts can be explained by four different theories.3,6,7,11) The dysraphic theory claims that cysts are formed as a result of a pause in the development of the neural tube or the surrounding mesoderm during embryogenesis.14) The acquired dural rupture theory blames direct trauma, defined as the deprivation of the integrity of the dura during surgical interventions, and indicates that the cysts are caused by lumbar puncture needles and hernia of the arachnoid.7) Our third case can be explained by this theory because the complaints started after trauma. The proliferative theory was introduced after the analysis of myeloschisis pathogenesis, and
Intrasacral Extradural Arachnoid Cyst

Table 1  Classification of spinal meningeal cysts

<table>
<thead>
<tr>
<th>Type</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ia</td>
<td>extradural meningeal cyst without spinal nerve root fibers</td>
</tr>
<tr>
<td>Ib</td>
<td>sacral meningocele (occult sacral meningocele)</td>
</tr>
<tr>
<td>I</td>
<td>extradural meningeal cyst with spinal nerve root fibers (Tarlov’s perineurial cyst, spinal nerve root diverticulum)</td>
</tr>
<tr>
<td>II</td>
<td>spinal intradural meningeal cyst (intradural arachnoid cyst)</td>
</tr>
</tbody>
</table>

suggests that the closing of the neural tube was prevented by local enlargement. The final theory is insufficiency of the exit of the meningeal sac to the cranium introduced in 1932. The neural tube and the dural sac are extended to the coccyx in filum caudal form, then the connection between the neural tube and the filum caudal is terminated, and the neural tube retracted towards the cranium. The connection to this point of the initiation continues as filum terminale. In adults, the dural sac is contracted around the filum terminale. In individuals without such migratory movement, a sacral cyst might be formed.

Intrasacral extradural cysts generally become symptomatic in the 3rd or 4th decade of life, as in our patients. Although cases are mostly asymptomatic, the most important sign in symptomatic cases is intermittent and chronic blunt pain in the lumbar region and perinea, encountered in 70% of cases. Gluteal neuralgia was reported in one case. Radiculopathy may be present. Pain increases with physical activity and the Valsalva maneuver or standing up.

Our cases had similar presentations. Clinical symptoms and findings may give the impression of lumbar disc hernia, but lumbar disc hernias and intrasacral extradural arachnoid cysts can occur together. Under these circumstances, the origins of the symptoms must be differentiated.

In our first case, lumbar disc hernia was encountered, but the clinical symptoms and findings were related to the intrasacral extradural arachnoid cyst. Perineal paresthesia and sexual impotence have also been reported in some patients. Although sensory symptoms are present, motor weakness is rare in the lower extremities. Sphincter malfunctions such as dysuria, retention, constipation, incontinence, and repetitive urinary system infections can also be encountered.

With the exception of our first patient, who had no Achilles reflex, all other patients appeared normal during neurological inspection. However, lumbosacral sensitivity was present in all patients. The presence of skin lesions, that may be a symptom of dysraphism, has not been reported with intrasacral extradural arachnoid cysts, as in our cases. Association with tethered cord syndrome as a result of thick filum terminale has been reported.

Intrasacral extradural cysts were encountered at a higher rate in females than in males.

MR imaging is the best diagnostic tool for detection of intrasacral extradural arachnoid cyst, and has resulted in an increasing number of incidentally diagnosed cases. MR imaging should involve with a minimum of T₁, T₂, and proton density-weighted imaging and administration of contrast medium. Both T₁-weighted imaging are essential to define the wall and the contents of the cyst. Both T₂-weighted MR imaging show a cystic lesion as isointense to cerebrospinal fluid.

In our patients, MR imaging of the lumbosacral region revealed dilated cystic lesion in the sacral spinal canal.

Intrasacral extradural arachnoid cysts become symptomatic due to pressure to the nerve roots, resulting in radicular complaints. The mechanism of these radicular symptoms can be attributed to compression and/or displacement of the nerve roots due to the space-occupying effect of the cyst. Prompt surgical intervention is necessary to prevent permanent damage to the nerve roots. The surgical approach is via sacral laminectomy, which usually reveals thin laminae. Total excision of the cyst is unnecessary, and obliteration of the fistulous tract is the optimal treatment. Other surgical options are possible, such as simple bony decompression via sacral laminectomy, cyst resection, incision and drainage of the cyst, plication of the cyst, lumboperitoneal cerebrospinal fluid shunting, and endoscopic closure of the communication of the cyst with the dural sac.

In both our cases, resection of the cyst and primary closure of its connection with dural sac were sufficient.

The cyst consists of fibrous tissue resembling dura mater that is usually lined by arachnoid membrane and lies within the enlarged sacral spinal canal. The presenting symptoms reflect local compression of the sacrococcygeal roots and adjacent structures. Therefore, postoperative resolution of pain and paresthesia occur in the vast majority of cases after decompression of cystic lesions.

Our patients showed marked improvement or resolution of pain and paresthesia in the immediate postoperative period.

In conclusion, intrasacral extradural arachnoid cysts should be considered in the differential diagnosis of pain in the low back and perineal region. MR imaging is the best method of confirming the diagnosis and surgery is curative.
References


Address reprint requests to: Mustafa Kemal Hamamcioglu, M.D., Department of Neurosurgery, Trakya University Faculty of Medicine, Posta Kutusu: 23, Edirne 22001, Turkey.

*Tel*: +90 262 422 23 12

*e-mail*: mkemalh2@yahoo.com

*mkemalh@hotmail.com*