Recurrent Lumbar Spinal Intradural Enterogenous Cyst: A Case Report

Rekürran Lomber Spinal İnadural Enterogenoz Kist: Olgu Sunumu

ABSTRACT

Enterogenous cysts mostly present in the cervical and thoracic region of the spinal canal and have only rarely been observed in the lumbar region. We report here a case of a recurrent enterogenous cyst in the lumbar spinal canal. A 24-year-old woman presented with lower back and left radicular leg pain for 1 year. She had been operated on for mass lesion at the same level 10 years ago. Lumbar spine MRI showed a large intradural cyst at L2. Other congenital abnormalities were absent. The cyst was subtotally removed and the patient recovered well. Pathological examination revealed that the lesion was a typical enterogenous cyst.

KEYWORDS: Enterogenous cyst, Neuroenteric cyst, Spinal cord

ÖZ


ANAHTAR SÖZCİKLER: Enterogenoz kist, Nöroenterik kist, Spinal cord

1,2,3,4,5 School of Medicine, Karadeniz Technical University, Department of Neurosurgery, Trabzon, Turkey
6 School of Medicine, Karadeniz Technical University, Department of Pediatrics, Trabzon, Turkey
7 School of Medicine, Karadeniz Technical University, Department of Pathology, Trabzon, Turkey

Received : 17.03.2009
Accepted : 31.07.2009

Correspondence address:
Erhan ARSLAN
Department of Neurosurgery, School of Medicine, Karadeniz Technical University, 61080 Trabzon, TURKEY
Phone : +90 462 377 52 53
Fax : +90 462 322 10 21
E-mail : arserhan@gmail.com
INTRODUCTION

The term enterogenous cysts was used for the first time by Harriman to describe cysts previously known as neuroenteric, endodermal or respiratory cysts, (7). Enterogenous cysts typically present with spinal cord or cranial nerve compression and are rare, nonneoplastic intradural mass lesions occurring in childhood or early adulthood (5,9). Vertebral and spinal cord abnormalities are frequently seen in cases of cysts involving the spinal canal, and these cysts may communicate with extraspinal cysts anterior to the spine (1,13). The cysts are typically lined by columnar mucin-producing epithelium resembling enteric epithelium or columnar ciliated epithelium resembling respiratory epithelium, and are usually 1 cm or less in size. They are rarely lined by nonkeratinizing squamous epithelium or by stratified cuboidal epithelium (13,15). Enterogenous cysts have previously been reported in the spinal canal (1). Cervical and upper thoracic segments are most often affected, and these cysts are generally very rare within the lumbar spinal canal.

CASE REPORT

A 24-year-old woman complained of lower back and left radicular leg pain for 1 year. She had first felt lower extremity weakness with numbness several months before admission. Her symptoms became progressively worse and she eventually experienced difficulty walking. She had been operated upon for a mass lesion at the same level 10 years ago. The cyst wall had ruptured and could not be resected because of the technical inadequacy of the operating microscope and surgical devices at the time and an exact histopathological diagnosis had therefore not been made.

On admission, physical examination revealed left lower extremity grade 2 motor weakness and right-side grade 1 to 2 motor weakness. Reflexes were normal in the legs. She had hypoesthesia in the L1, L2, and L3 dermatomes in both legs. Lumbar spine MRI images showed a low conus medullaris due to a tethered cord with a hyperintense cystic mass in the medulla spinalis at the level of L2 (Figure 1A, B).

After dissection of the paravertebral muscles, no laminectomy was needed. A cystic mass approximately 3 cm in diameter was found when the dura was opened along the posterior midline. The cystic mass was easily dissected away from the neural tissues with posterior midline myelotomy. The viscous content of the cyst was evacuated through needle aspiration, and the collapsed wall was dissected and removed as completely as possible. Histopathology (Figure 2) showed a thin fibrous cyst wall lined by a mucus-secreting columnar epithelium, which is consistent with an enterogenous cyst.

The postoperative course was uneventful and the lower back and left leg pain improved immediately. Muscle strength had returned to normal three months later. The patient continued to do well 8 months later.

Figure 1: Sagittal T2-weighted MRI of the lumbar spine shows (A) a large cystic lesion at L2 filling almost all of the entire spinal canal. Previously created L1, L2 and L3 laminectomy defects are seen. Low conus medullaris at the level of L3-4 is also seen with tethered cord. (B) In the gadolinium enhanced sagittal T1-weighted image, there is no enhancement of the cyst.

Figure 2: Microscopic cross-section examination demonstrates a cyst wall lined by mucin-secreting columnar epithelium (hematoxylin & eosin staining, magnification x400).
DISCUSSION

Enterogenous cysts are considered congenital anomalies. General localizations of these cysts are in the mediastinum, abdominal cavity, and the spinal canal or skull (22). Enterogenous cysts that occur in and around the central nervous system (CNS) are rare lesions of presumed endodermal origin. Endodermal, neuroenteric, epithelial, bronchogenic, respiratory, and foregut cyst are the other names that have been used for these types of lesions (23). The general location for the intraspinal enterogenous cysts is in the subarachnoid space anterior to the spinal cord (22).

Enterogenous cysts of the CNS are lesions of controversial nomenclature, pathogenesis, and classification (4,5,9,18). Some authors have cited their associations with vertebral and spinal cord abnormalities as evidence of origin from displaced embryonic endodermal elements (1,10). Others, citing the midline location, have suggested a teratomatous origin (8). Ciliated epithelium and mucinous epithelium, both potentially of endodermal origin, are observed in enterogenous cysts. Although the origin of the cyst epithelium is controversial, an endodermal origin is currently supported rather than neuroectodermal origin based on the presence of mucin-producing epithelium in some cases (15).

The ventral cervicothoracic spinal canal is the usual location for these cysts; however, they may be found in the cervical spine, the lumbar spine, and even in the cranium (4,12,13,19,21). They are usually seen in the intradural extramedullary location, but are also rarely found in the intramedullary compartment (2,16,24). These cysts are located posterior to the spinal cord in a small number of patients (25). These cysts may be purely intraspinal, as with the patient reported in this article, mediastinal, or involve malformations of both (14,20). The dual malformation cysts are usually connected through an adjacent vertebral defect called the Kovalesky canal (6,11,14,17). Hemivertebrae, unsegmentation of vertebra, spina bifida, and clefts in the vertebral bodies are all vertebral abnormalities that are seen in this condition. Spinal cord anomalies that include diastematomyelia and split cord malformation are frequently associated with enterogenous cysts (3). There were neither spinal cord anomalies nor vertebral abnormalities in the patient we reported here.

Neural decompression and prevention of cyst refilling are the aims of surgical treatment (3). Radical removal of extramedullary intraspinal cysts that are causing spinal cord compression and drainage of the cystic lesion represent the surgical therapy of choice (11,14). Total resection of the intramedullary component is generally not advocated as there is a high risk of damaging the spinal cord and operative interventions have ranged from cyst aspiration to subtotal resection with cyst marsupialization (2,16). The cyst wall was resected subtotally after aspiration of the cyst’s content with a needle in our case.

CONCLUSIONS

Total resection of the intramedullary component of these cysts is generally not advocated because of the high risk of damaging the spinal cord. Recurrence of these intraspinal enterogenous cysts after cyst aspiration and subtotal resection is therefore the biggest problem after surgery.

REFERENCES