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Ventral Intradural Endodermal Cyst in the Cervical Spine Treated With Anterior Corpectomy

—Case Report—

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Abstract

A 14-year-old girl who presented with an endodermal cyst manifesting as severe neck and shoulder pain along with vesicorectal disturbances. Cervical magnetic resonance imaging showed a slightly enhanced intradural cyst at the C6-7 level in the ventral side of the spinal canal, with significant dorsal shortening and thinning of the spinal cord. Anterior corpectomy was chosen because of the dorsal effacement of the spinal cord. The cyst wall was subtotally removed to avoid damage to the normal spinal cord. After cyst removal, the iliac bone and an anterior cervical plate were used for anterior fusion. Postoperatively, her pain subsided without neurological deficits. The histological diagnosis was endodermal cyst. The cyst did not recur during a follow-up period of 18 months. Endodermal cysts are rare congenital lesions of the spine lined by endodermal epithelium. The natural history of this lesion is unclear, and the surgical strategy for the approach route and the extent of removal of the cyst wall remain controversial. We suggest that the anterior approach may allow a safer and more effective surgical route for the treatment of ventrally located endodermal cyst compared to the posterior approach.

Key words: endodermal cyst, neurenteric cyst, recurrence, spine, surgical approach

Introduction

Spinal endodermal cyst is a rare congenital lesion histologically characterized by epithelium consisting of mucin-producing columnar cells and underlying connective tissue.[24] Endodermal cyst is also called neurenteric cyst. Such cysts are generally located ventral to the spinal cord, and were mainly treated surgically via an anterior[2,3,5,6,8,9,11,14,16,19] or posterior approach.[1,4,13,15,17,18,20,21,23] However, these techniques have not been directly compared, and the surgical indications and strategy for such cysts remain controversial. We describe a case of cervical ventral endodermal cyst treated successfully by anterior corpectomy and discuss the associated benefits of this approach.

Case Report

A 14-year-old girl presented with a 2-month history of neck and shoulder pain associated with vesicorectal disturbances. The shoulder pain was progressive, and she could not raise either arm because of severe pain at presentation. Cervical magnetic resonance (MR) imaging at a local clinic showed a cyst at the C6-7 level, and she was referred to our department for further examination and treatment.

On admission, she complained of severe neck pain projecting to the bilateral shoulders, which decreased at rest. She had no motor weakness, but hyperreflex of the left biceps and patella tendon were noted. Cervical MR imaging demonstrated slightly enhanced intradural cyst measuring 11 × 18 × 23 mm at the C6-7 level (Fig. 1). The cyst was positioned anteriorly, which caused significant symmetrical dorsal shortening and thinning of the spinal cord. Associated spinal malformations such as spina bifida, hemivertebrae, fused vertebral bodies, and Klippel-Feil anomaly were not identified.

The anterior approach was selected because of the dorsal effacement of the spinal cord. Under the operating microscope, corpectomy of C6 was performed using a high-speed drill. The inferior margin of C5 and the superior margin of C7 were also drilled out to obtain an adequate operative field. After gently removing the posterior longitudinal ligament, the dura mater was exposed. Longitudinal incision and reflection of the dura and arachnoid revealed the cystic lesion (Fig. 2). The cyst contained clear fluid, which was drained, and the visible ventral cyst wall was excised to the greatest possible extent. The dorsal wall was not well demarcated from the pial layer of the spinal cord, so we did not attempt resection, espe-

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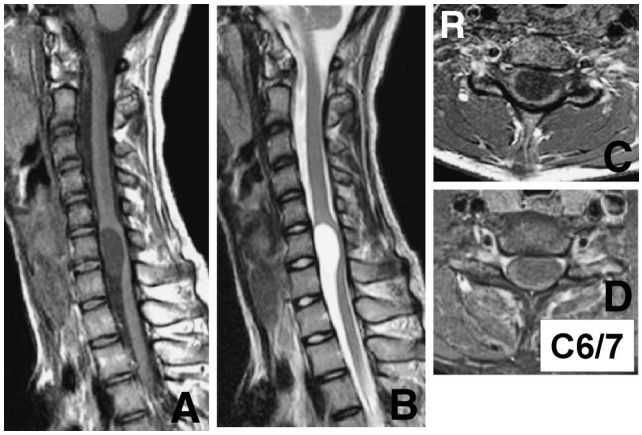

Fig. 1 A, B: Cervical sagittal magnetic resonance (MR) images showing the intradural cystic lesion at the C6-7 level appearing as low signal intensity on the T_1-weighted image (A) and high signal intensity on the T_2-weighted image (B). C: Cervical axial T_1-weighted MR image showing the intradural cystic lesion at the C6-7 level appearing as low signal intensity. The cyst is positioned anteriorly, and caused significant symmetrical dorsal shortening and thinning of the spinal cord. D: Cervical axial T_1-weighted MR image with gadolinium showing a slightly enhanced intradural cyst.

Fig. 2 Intraoperative photograph showing an endodermal cyst following reflection of the dural and arachnoid layers.

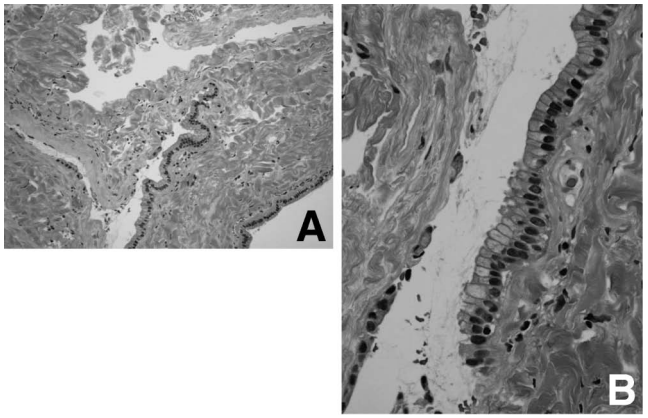

Fig. 3 Photomicrographs of the cyst wall showing a single layer of columnar, non-ciliated epithelium, suggesting the diagnosis of endodermal cyst. Hematoxylin and eosin stain, original magnification A: ×40, B: ×200.

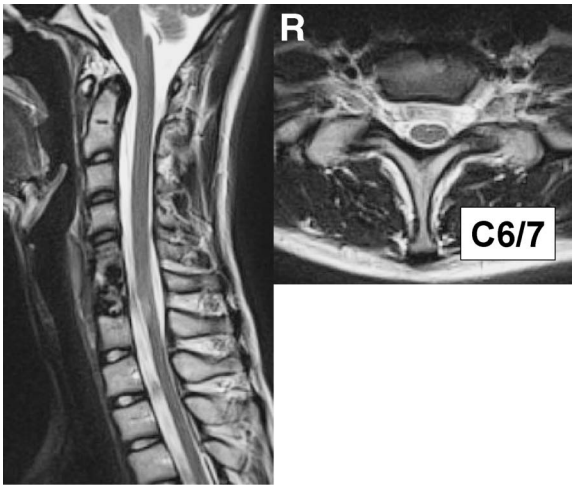

Fig. 4 Postoperative cervical T_1-weighted magnetic resonance images taken 18 months after operation showing no intradural cystic lesion at the C6-7 level. The spinal cord contour is restored after removal of the cyst.

cially in the rostral side of the cyst wall, as preoperative neuroimaging had shown that the cyst had encroached on the spinal cord, suggesting that the cyst was severely adhered to the spinal cord. The dura was approximated with Gore-Tex® sutures (W. L. Gore & Associates, Inc., Flagstaff, Arizona, USA). The approximated dura mater was covered with NEOVEIL® (Gunze Limited, Tokyo) to prevent cerebrospinal fluid leakage. Anterior spinal fusion was performed using harvested iliac bone and anterior titanium plate and screws.

After surgery, her severe neck and shoulder pain were immediately alleviated. Histological examination of the specimen showed a single layer of columnar, non-ciliated epithelium, suggesting the diagnosis of endodermal cyst (Fig. 3). Follow-up MR imaging performed 18 months after surgery showed no recurrence of the cyst and confirmed the expansion of the spinal cord (Fig. 4). Serial computed tomography scans demonstrated solid fusion of the bone graft. The anterior plate was removed 1.5 years after cyst removal. Postoperative spinal kyphosis did not occur during the follow-up period. Long-term follow up is needed to detect cyst recurrence.

Discussion

Endodermal cysts are rare developmental anomalies that may account for 0.01% of spinal tumors.[3,7] Dysgenesis of the endoderm with neurenteric canal formation at the notochord in the third week of embryogenesis has been proposed as an explanation of endodermal cyst formation.[23] Endodermal cysts are cystic lesions lined by cuboidal and columnar pseudostratified epithelium with ciliated cells resting on a base of connective tissue. The endodermal cyst is found at intracranial and intraspinal lo-

Table 1 Previous cases of spinal endodermal cyst treated with the anterior approach

Author (Year)	Age (yrs)/ Sex	Level	Symptoms	Procedure	Outcome	Complications	Recurrence
Arai et al. (1992)[5]	16/M	C5-6	paraparesis	corpectomy	good	CSF leakage	(−)
Devkota et al. (1994)[8]	6/M	C4-6	neck pain, paraparesis	corpectomy	good	(−)	(−)
Menezes and Ryken (1995)[14]	8/M	C4-6	recurrent meningitis	corpectomy	good	(−)	(−)
Fujimoto et al. (1996)[9]	15/M	C4-5	neck pain, monoparesis	corpectomy	good	(−)	(−)
Miyoshi et al. (1998)[16]	8/M	C6-T1	neck pain	corpectomy	good	(−)	(−)
	18/M	C6-7	neck pain	corpectomy	good	(−)	(−)
	9/M	C3-6	neck pain	corpectomy	good	(−)	(−)
	9/M	C7-T1	paraparesis	corpectomy	good	(−)	(−)
Agrillo et al. (2001)[2]	17/M	C2-4	paraplegia	corpectomy	good	(−)	(−)
Asazuma et al. (2002)[6]	24/F	cervical	neck pain	corpectomy	good	(−)	(−)
Oyama et al. (2004)[19]	46/F	C3-4	paraparesis	discectomy	good	(−)	(+)
Kusaka et al. (2005)[11]	28/M	C5-6	hemiparesis	corpectomy	good	(−)	(−)
Akil et al. (2009)[3]	15/M	T1-2	paraparesis	corpectomy	good	(−)	(−)
Present case	14/F	C5-6	neck pain	corpectomy	good	(−)	(−)

CSF: cerebrospinal fluid, F: female, M: male.

Table 2 Previous cases of spinal endodermal cyst treated with the posterior approach

Author (Year)	Age (yrs)/Sex	Level	Symptoms	Outcome	Complications	Recurrence
Takemi et al. (1984)[23]	12/F	C3	tetraparesis	good	(−)	(−)
Agnoli et al. (1984)[1]	39/F	C3-4	monoparesis	good	(−)	(−)
Matsushima et al. (1985)[13]	8/F	C5-T1	tetraparesis	good	(−)	(−)
Miyagi et al. (1988)[15]	11/F	C3-5	neck pain	good	(−)	(−)
Mizuno et al. (1988)[17]	6/M	C2-5	monoparesis	fair	chemical meningitis	(+)
Osenbach et al. (1992)[18]	32/F	C2-3	unknown	good	(−)	(−)
	7/M	C2-3	tetraparesis	good	(−)	(+)
Shenoy and Raja (2004)[21]	4/M	C2-3	tetraparesis	good	(−)	(−)
	3/M	C7-T1	paraparesis	poor	arachnoiditis	(−)
	16/F	C3-4	neck pain	good	(−)	(−)
	5/F	C6-8	paraparesis	good	(−)	(+)
Sheaufung et al. (2009)[20]	4/F	T1-2	paraparesis	good	(−)	(−)
Anan et al. (2010)[4]	5/F	C6-7	neck pain	good	(−)	(−)

F: female, M: male.

cations within the central nervous system.[19] Cysts in the intracranial region are extremely rare, generally located in the posterior fossa along the midline or in the cerebellopontine angle,[7] whereas cysts in the intraspinal region are more frequent, mainly located at the ventral aspect of the lower cervical and upper thoracic spinal cord. However, a few cysts have been located dorsal to the cord as well as in disseminated form.[24] Approximately 50% of intraspinal endodermal cysts are associated with spinal malformations, such as spina bifida, hemivertebrae, fused vertebral bodies, and Klippel-Feil anomaly.[12,22] The natural history of this lesion is unclear. No case of surgical resection in the asymptomatic lesion has been described. Patients with endodermal cysts presenting with neck pain and motor weakness are good candidates for surgical cyst resection.

Surgical indications and strategy for such cysts remain debatable. The standard treatment is drainage and removal of the extramedullary cyst component.[10] Radical resection of the spinal cord component of the cyst wall is not recommended as significant risk has been reported to be associated with this procedure.[17] In contrast, complete resection of the cystic wall adherent to the spinal cord has been recommended.[6] Therefore, the surgical strategy concerning the approach route and extent of removal of the cyst wall remains controversial.

Tables 1 and 2 show a summary of previous cases of endodermal cysts treated via the posterior[1,4,13,15,17,18,20,21,23] or anterior approach[2,3,5,6,8,9,11,14,16,19] including the present case. Three of 4 cases of recurrence of the cyst were treated via the posterior approach, and the other case was treated via the anterior approach though the intervertebral disc space. The interval between the initial surgery and recurrence was 1 to 13 years.[17–19,21] The anterior approach may provide an insufficient surgical field compared to that afforded by anterior corpectomy for adequate

removal of the cyst wall. If ventrally situated cysts are approached posteriorly, cyst resection is generally difficult without manipulating the spinal cord, even if the dentate ligament is divided and used to rotate the cord. One of the technical problems of the posterior approach is sudden shrinking of the cyst wall following partial excision. Extensive spinal cord retraction is needed for further excision of the cyst, especially for manipulation of the cyst wall adherent to the spinal cord. These factors make adequate resection difficult to achieve. Dissemination of the cyst in the entire spinal cord is also possible.[24] Therefore, we consider that excise of the cyst wall to the maximum possible extent is important to prevent cyst recurrence and dissemination.

The anterior approach is technically complex and requires subsequent spinal fusion, but provides direct and good visualization of the cyst, and allows safe removal of the cyst wall. No severe surgery-related complications associated with the anterior approach have been reported.[2,3,5,6,8,9,11,14,16,19] Thus, the anterior approach is safe for reaching a cyst located ventral to the spinal cord, as the relationship between the cyst wall and the spinal cord can be clearly visualized. Moreover, although adequate removal of the cyst wall of endodermal cysts is difficult to perform in the deep, narrow operative field that is obtained in the discectomy approach, this is not hold true for corpectomy. A wide operative field for confirmation of removal of the whole cyst can be obtained via anterior corpectomy using an iliac bone graft because of the cyst location.

We consider that the anterior approach in this case allowed safer and more effective treatment of the endodermal cyst, and we believe that anterior corpectomy is a suitable approach for treating ventrally located cervical intradural lesions based on our experience and previous reports.

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