

Case Report

Anterior cervical intradural arachnoid cyst in a child: a case report and literature review

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Abstract: Intradural arachnoid cyst is classified as type III meningeal cyst according to the histological examination. These are most commonly found in the posterior thoracic spine, while those in the anterior cervical spine are extremely rare. We report our experience with a case of the anterior cervical intradural arachnoid cyst. Key findings from a literature review are presented and comparisons drawn with the previously reported 30 cases. A 7-year-old boy presented with a history of neck pain and difficulty in walking. Magnetic Resonance Imaging (MRI) examination showed an anterior cervical intradural arachnoid cyst, which had caused spinal cord compression and deformation. The child received laminectomy combined with cystectomy. The symptoms significantly improved after surgery. However, follow-up MRI six months after surgery showed recurrence of the arachnoid cyst pressing down on his spinal cord. MRI offers a definitive advantage in evaluation of this disease. Postoperative recurrence is common irrespective of the surgical procedure. Surgery is still the most effective method for treatment.

Keywords: Cervical, intradural, anterior, arachnoid cyst, child

Introduction

Anterior cervical intradural arachnoid cyst (Type III meningeal cysts) is a rare clinical entity [1]. About 80% of these lesions are found in the thoracic spine in a posterior location, while only a few appear in the cervical spine [2, 3]. Anterior cervical intradural arachnoid cysts are extremely rare; only 30 cases have been reported till date [2, 4-27]. Most of the reported cases presented with neck pain and symptoms, caused by spinal cord compression, such as muscle weakness and sensory disturbances. Most of these cases have been treated by laminectomy and cyst fenestration via a posterior approach; only five cases have been treated via anterior cervical approach (corpectomy, and excision of the lesion). In this paper, we present our experience with a 7-year-old boy with an anterior cervical intradural arachnoid cyst; he was operated with complete neurological recovery. However, MRI examination performed after six months revealed recurrence of the cyst. Key findings of a review of relevant literature are presented along with discussion on the etiopathogenesis, clinical presenta-

tion, imaging characteristics and treatment of intradural arachnoid cyst (**Table 1**).

Case report

A 7-year-old boy was admitted with chief complaints of neck pain since the last six months and difficulty in walking since 15 days ago. The patient had a history of intermittent episodes of neck pain. The pain was not relieved after rest but only by painkiller. The patient exhibited difficulty in walking and had sustained pain in the right ankle that was not relieved by oral analgesics. There was no history of change in temperament, convulsions, abdominal pain or distension, palpitation, or chest pain. Ankle X-ray demonstrated no obvious abnormalities. MRI findings showed a uniform signal in the spinal canal at cervical 4, 5 level with low signal in T1WI and high signal in T2WI which were consistent with cerebrospinal fluid. The spinal cord was compressed, deformed, and displaced; however, there was no evidence of bone destruction (**Figure 1A-D**). The patient had no history of meningitis or trauma. Physical examination demonstrated normal development and

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Table 1. Cases of anterior cervical intradural arachnoid cyst reported in literature

Year	Author	Age/ Sex	Patient history	Symptom	Affected spinal level	Treatment	Outcome
1960	Hoffman	6/M	Not clear	Paraplegia unspecified	C2-C3	Laminectomy	Improve
1974	Palmer	19/F	Not clear	Flaccid tetraplegia	C1-C3	Laminectomy, complete cyst resection	Improve
		3/M	Not clear	Pneumonia	C2-C4	Laminectomy, multiple needle punctures	Die
1978	Duncan	3/F	Not clear	Neck pain, hemiparesis	C0-C2	Laminectomy, cystectomy	Improve
1978	Herskowitz	28/F	Not clear	Flaccid tetraplegia	C6-C7	Laminectomy, cystectomy	Improve
1982	Kendall	31/F	Not clear	Spastic paraplegia	C5-C7	Laminectomy, cystectomy	No improve
1985	Chan	37/F	Not clear	Upper extremity strength decrease	C1-T5	Laminectomy, cyst-laparotomy	Improve
1990	Jena	17/M	Not clear	Flaccid tetraplegia	C7-T1	Laminectomy, cystectomy	Improve
1992	Rabb	2/F	Not clear	Meningomyelocele	C6-C7	Laminectomy, cyst-thoracic shunt	Improve
1996	Chen	18/M	C2 fracture for 9 years	Left side hemiparesis	C3-C5	Laminectomy, cystectomy	Improve
1998	Jean	14/F	Meningomyelocele, Chiari II malformation	Headache, swirl	C0-C6	Cyst-peritoneal shunt	Improve
		9/M	Meningomyelocele, Chiari II malformation	Quadriplegia, neck pain	C0-C5	Laminectomy, cyst suction	No improve
1999	Kazan	18/M	Cervical spinal trauma for 2 months	Quadriplegia, neck pain	C6-C7	Laminectomy, cystectomy	Not clear
		15/M	Cervical spinal trauma for 2 weeks	Quadriplegia	C2-C3	Partial laminectomy with open window for cystectomy	Improve
2001	Lee	4/M	Not clear	Paraparesis	C5-T1	Laminectomy, cystectomy	Improve
2002	Safrie	26/M	Not clear	Left side hemiparesis	C5-T3	Laminectomy, cystectomy	Improve
2003	Takahashi	13/M	C1 spina bifida occulta	Severe headache	C1-C3	MRI guidance percutaneous puncture	Improve
2003	Banczerowski	22/M	Not clear	Neck pain, upper extremity palsy	C6-C7	Anterior laminectomy, cystectomy	Improve
2004	Muthukumar	4/M	Trauma (fall injury)	TLLOC, limbs paraplegia, tendon reflexes diminished	C3-C4	Laminectomy, cystectomy	Improve
		14/F	Trauma (fall injury)	Walking unsteady	C6-C7	Laminectomy, cystectomy	Improve
2006	Maiuri	43/F	Not clear	Quadriplegia	C7	Laminectomy, windowing, cystectomy	Improve
2008	Gezici and Ergun	2.5/M	Meningitis	Neck pain, quadriplegia	C7	Laminectomy, windowing, cystectomy	Improve
2008	Muhammedrezai	29/M	Not clear	Lower limb paralysis	C6	Anterior C7 laminectomy, cystectomy	Improve
2008	Campos	5/F	Not clear	Neck pain, upper extremity palsy	C1-C2	Laminectomy, cystectomy	Improve
2009	Srinivasan	51/M	Not clear	Neck pain, right upper extremity palsy	C2-C3	Anterior center C2 laminectomy	Die
2012	Rahimizadeh	2/F	Torticollis	Neck pain, left upper extremity paralysis	C5-C6	Laminectomy, cystectomy	Improve
		17/M	Torticollis	Neck pain, quadriplegia	C2-C3	Laminectomy, cystectomy	Improve
2016	Engelhardt	18/M	Trauma (traffic accident)	Neck pain, quadriplegia	C2-C5	Anterolateral C5 laminectomy	Improve
2016	Demir	23/M	Neck trauma	Neck, back, and arm pain	C3-T1	Laminectomy, cystectomy	Improve
2017	Shrestha	20/M	Not clear	Neck pain, quadriplegia	C3-C4	Ventro-center C3-C4 laminectomy	Improve
2017	This study	7/M	Not clear	Neck pain, quadriplegia, limb paralysis	C4-C5	Cystectomy	Improve

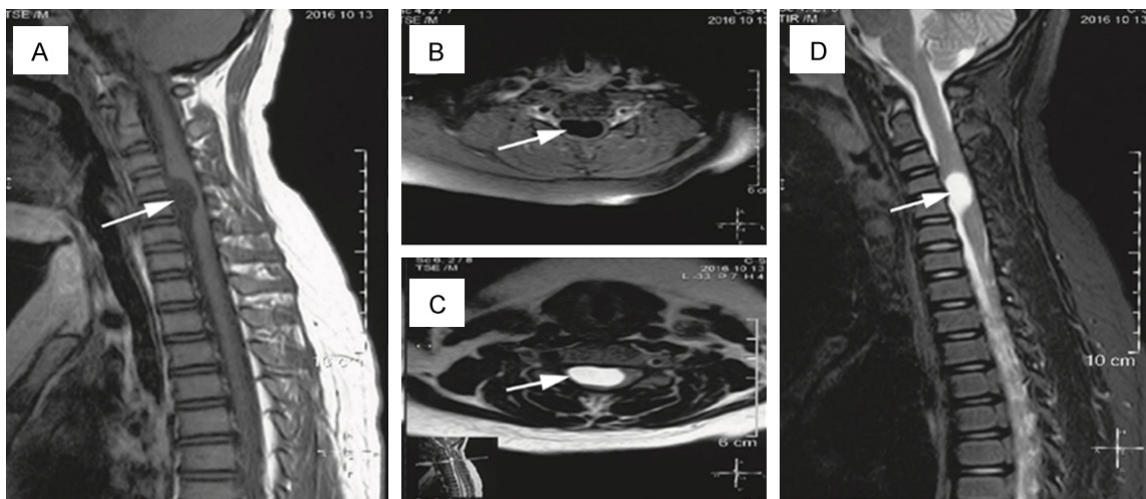


Figure 1. Preoperative MRI examination of cervical spine. A. Sagittal scanning with SE sequence (T1WI) exhibits oval signal of uniform density, and has a clear boundary with smooth margins. B. Coronal scanning with SE sequence (T1WI) exhibits a low signal. C. Coronal scanning with GRE sequence (T2WI) showed a high signal. D. Sagittal scanning with GRE sequence (T2WI) showed that posterior spinal cord is compressed and deformed.

normal sensorium. Pupils were round, bilaterally equal (diameter 3.0 mm) and reactive to light. Right limb muscle strength was grade IV, while the left limb muscle strength and muscle tension were normal. Physiological reflexes were normal; and no pathological reflex was found.

Intraoperatively, the spinal cord was found to have expanded towards the rear. The arachnoid on the spinal cord surface was carefully incised to release the cerebrospinal fluid. The spinal cord tension obviously decreased. Subsequently, the spinal cord was gently pulled to the side and the ventral side of the spinal cord explored through the space between cervical 4 and 5 nerve roots on the right side. A gray-colored cystic space-occupying lesion was observed. A total of 2 mL colorless transparent capsule fluid was extracted. No significant adhesion between the cyst wall and the spinal cord was found. The cyst wall was completely resected under an electron microscope and electrophysiological monitoring.

Postoperatively, there was a marked relief of neck pain with full recovery of muscle strength. The right foot discomfort significantly reduced. On histopathological examination of surgical specimen, the cyst wall was found to be composed of elastic fibers (**Figure 2A, 2B**). And the diagnosis of arachnoid cyst was made. MRI examination performed six months after sur-

gery revealed the cyst was still present (**Figure 3A, 3B**). We suspect that the effect of the chronic stress on spinal cord morphology led to recurrence of cyst. The patient's symptoms have relieved, so that we tell the patient to perform MRI examination after 1 year. If the condition worsens, visit the doctor in time please.

Discussion

In this study, we found that most cysts in the cervical spinal canal are of idiopathic etiology. In clinical settings, neck pain and extremity dyskinesia are the most common presenting symptoms. MRI has a certain diagnostic value based on its specific characteristic. Surgery is the treatment of choice for this disease.

The etiopathogenesis of spinal canal cyst is not completely understood. The lesion can be subdural or epidural in location. In 1988, Nabors categorized spinal canal arachnoid cysts into three types based on surgical and histological characteristics. Type I refers to epidural arachnoid cyst which does not involve the spinal nerve roots; type II is epidural arachnoid cyst which involves the spinal nerve roots; while type III is the intradural arachnoid cyst [1]. Type III arachnoid cyst can be further divided into congenital and idiopathic; the present case is likely to be that of congenital arachnoid cyst. Congenital subdural arachnoid cyst is formed

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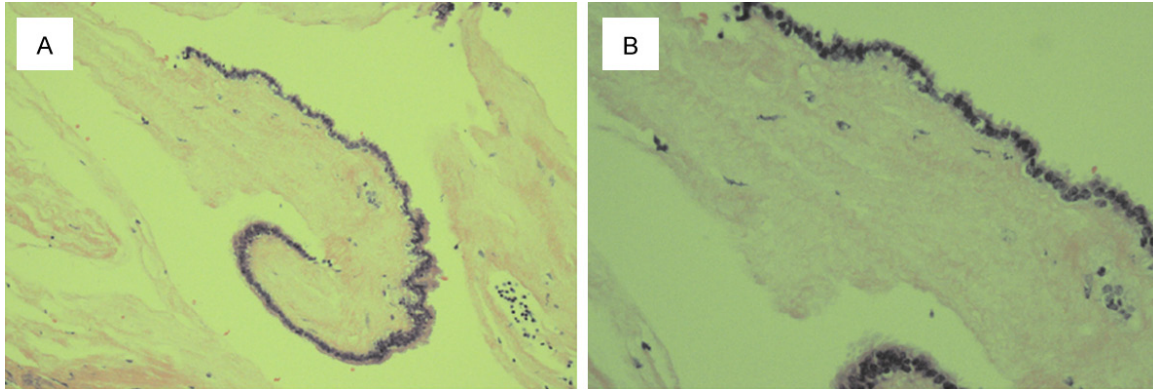


Figure 2. Histological findings from hematoxylin and eosin staining. Histological examination revealed fibrous connective tissue in the cyst wall (HE, $\times 20$, $\times 40$).

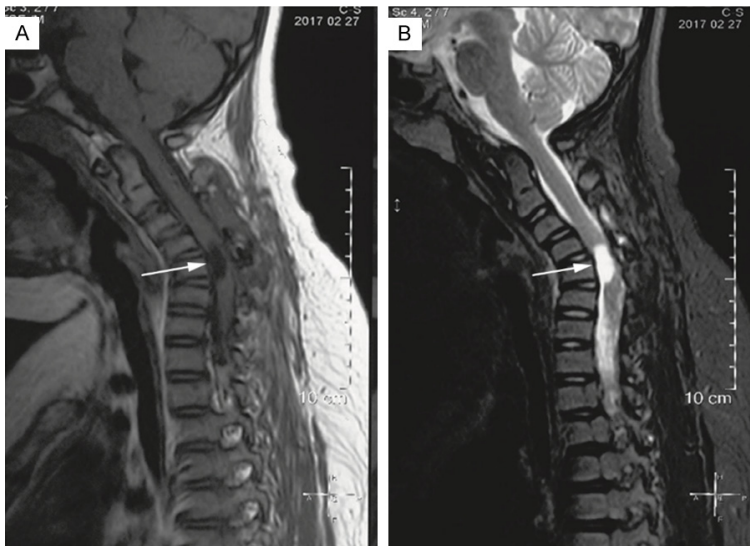


Figure 3. Postoperative MRI examination of cervical spine. A. Sagittal scanning with SE sequence (T1WI) exhibits a low signal, indicative of cyst recurrence. B. Sagittal scanning with GRE sequence (T2WI) showed a high signal and spinal cord is still deformed.

by cerebrospinal fluid flow obstruction because of abnormal proliferation of the arachnoid rhabdoliis. Primary arachnoid cyst in children is also associated with neural tube defects [14]. Idiopathic type III arachnoid cysts are often caused by arachnoid adhesions secondary to trauma, infection, surgery, or cancer [28]. Among the reported cases, 6 cases were of traumatic etiology; 3 cases were attributed to spinal cord disease, 2 cases to torticollis, and 1 case to meningitis.

The imaging characteristics are similar to those of cerebrospinal fluid. The cyst appears as a low density mass with clear boundary and

shows no enhancement on CT scan. MRI is the most preferred imaging modality for diagnosis of intradural arachnoid cyst [29]. MRI can help identify the location and contents of the cyst, and detect spinal cord atrophy and softening, which helps to predict the postoperative complications [30]. The ventral cervical spine intradural arachnoid cyst often exhibits oval or spindle signal of uniform density, and has a clear boundary with smooth margins. In the present case, T1WI showed low signal, while T2WI showed high signal. The capsule was not enhanced, which was consistent with cerebrospinal fluid. The differential diagnosis is from other common congenital cystic lesions, such as neuroepithelial cyst, intestinal cyst, and teratoid cyst.

Of the 33 reported patients, there were 22 males and 11 females; mean age of patients at diagnosis was 15.7 (2-51) years. Larger cysts or presence of nerve root compression may present with clinical symptoms related to the site of compression. Spinal canal arachnoid cysts show a chronic progressive growth process with the accompanying mass effect. It may compress the spinal cord or nerve roots with different clinical manifestations. The most common clinical manifestations are weakened muscle power (28 cases), neck pain (12 cases),

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deep and shallow hypesthesia or dysaesthesia. Urinary incontinence was reported in a few cases; headache caused by cervical cyst or angina caused by thoracic cyst were also uncommon [3, 20, 31, 32].

Operation is the most effective way to treat the disease [5, 33]. The main purpose of surgery is complete resection of the cyst to widen the CSF spaces posterior to the cord [30, 34, 35]. Surgical treatment includes laminectomy or laminoplasty, cyst-peritoneal shunt and cystectomy [36, 37]. Some scholars also suggested cyst puncture under CT or MRI guidance [38]. This method is less traumatic but is associated with frequent relapse. In a previously reported case series, 26 cases showed improvement in symptoms postoperatively, 2 cases died, and 2 cases exhibited no symptom remission. In this research, we found that most cases underwent cystectomy. If the spinal cord is compressed for a long time, which will lead to peripheral arachnoid thickening adhesions, the operators should be careful to separate the adhesion of the arachnoid and remove the cysts totally as much as possible. Most cases experienced postoperative cyst recurrence, the symptoms were obviously relieved. But in order to obviate the need for cord retraction, anterior approach through cervical body vertebrectomy has been suggested and done in five separate reports. The earlier the treatment, the better was the outcome.

Conclusion

Anterior cervical intradural arachnoid cyst is extremely rare. MRI offers a particular advantage in this disease. Postoperative recurrence is common irrespective of the surgical procedure. Operation is still the most effective method for the treatment. Postoperative spinal abnormalities were not significantly improved, possibly due to a prolonged compression of the spinal cord. Early detection and treatment can significantly improve clinical symptoms.

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Disclosure of conflict of interest

None.

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