Recurrent cervicodorsal spinal intradural enterogenous cyst: case report and literature review

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Abstract: This report described a recurrent enterogenous cyst of the cervicodorsal spinal canal occurred in an 8-year-old boy who experienced cervical back pain at the age of 5. He had been operated for mass lesion at the same level 3 years ago. The cervical and thoracic spine MRI showed a large intradural cyst at C7-T1. The cyst was subtotally removed via posterior approach using a laminectomy. Based on the results of immunostaining, it was identified as an enterogenous cyst. A literature review related to spinal cyst is also included.

Keywords: Recurrent enterogenous cyst, cervical spinal canal

Introduction

The cause of cervical spinal cord compression consists of cervical disc protruding, traumatic cervical injury, cervical spine instability, and spinal canal mass lesion [1]. Several studies have demonstrated that intraspinal cyst usually affects young patient with cervical spinal mass lesion [2-6]. Embryologically, the cyst is suggested to be related to congenital maldevelopment of endodermal tissue in the spinal canal and the neural plaque [2, 7], thus, the endodermal cysts should be considered in the diagnosis of spinal solitary cystic mass. It is known that the endodermal cysts usually include the enterogenous and neuroepithelial cyst, and develop in the subdural space in the anterior spinal cord [8]. Because an endodermal cyst wall includes columnar epithelium, its layer and its cells have the ability to grow and produce secretions, the cyst should be subtotally excised [9]. We report here a case of a recurrent enterogenous cyst in the cervicodorsal spinal canal.

Case report

An 8-year old boy presented with cervical back pain for 1 week. His nocturnal pain symptom became progressively worse. He didn’t experience weakness and paraesthesia of double upper extremity. He had been operated for mass lesion at the cervicothoracic spine level 3 years ago, and a postoperative diagnosis of enterogenous cyst was made. A MRI film of the cervicothoracic spine in November 9, 2014 showed a cystic mass lesion ventral to the spinal cord and lying at the level from C7 to T1 (Figure 1A and 1B). Written informed consent was obtained from the patient and his family before preparation and submission of this manuscript.

The patient underwent preoperative evaluation using a standard protocol. The following parameters were recorded during surgery: electrocardiogram, non-invasive blood pressure, pulse oximetry, body temperature, inspired and expired anesthetic concentration. In the operating room, after he was lightly sedated with 1 mg intravenous midazolam, anesthesia was induced with intravenous fentanyl 2 μg/kg, propofol 2.5 mg/kg. Intravenous administration of rocuronium 0.8 mg/kg was used to facilitate endotracheal intubation with a 5.0-mm-ID endotracheal tube (Cormack and Lehane grade I). 1%-1.5% isoflurane and 0.1-0.2 mg/kg/min propofol were used to maintain general anesthesia and PaCO$_2$ between 35-40 mmHg [10-17]. Intermittent doses of vecuronium were
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Under general anesthesia, the boy was placed in prone position with the neck extended. He was subject-
ed to operate on via poste-
rior approach using a C6-T1
laminectomy. After opening
the dura, the spinal cord
and a well-demarcated en-
capsulated cyst were expo-
sed by blunt dissection. It
was observed that the cyst
carried no obvious com-
pression to spinal cord. The
cyst was fragile and was
removed along its axis as
completely as possible and
excised from their attach-
ments using microscope.
The operation lasted 5 h.
After confirmation of neu-
romuscular recovery, the
patient was extubated in
supine position and trans-
ported to the post-anesthe-
sia care unit (PACU). The
patient was monitored for 1
h in the PACU. There were
no complications. After he
was conscious and his vital
signs were stable, he was
discharged to the ward. The
preoperative pain symp-
toms disappeared 5 days
after surgery. No additional
signs related to the cervical
nerves were observed. Pat-
hologically, the mass lesion
was diagnosed as an en-
terogenous cyst (Figure 2).
With the aid of postopera-
tive magnetic resonance
imaging, the mass lesion
within the C7-T1 spinal cord
was removed (Figure 1C
and 1D). The patient was
discharged and followed
up for 4 month, with no
symptoms.

Discussion

The neurenteric cyst usually located intradural-
ly or extramedullary in the ventral cervicodorsal

administered to maintain skeletal muscle
paralysis [18, 19]. The depth of anesthesia was
monitored by intraoperative recording bispec-
tral index (BIS) [20-22].

Figure 1. Composite photographs of preoperative (A, B) and postoperative (C, D) MRI images in sagittal view (left) and axial view (right). Sagittal T2-weighted MRI of the cervical spine (A) showed a large cystic lesion at C7-T1 filling almost all of the entire spinal canal. The C7-T1 spinal cord has been displaced posteriorly by enterogenous cysts (blue arrows). 6 d after surgery, sagittal T1-weighted sequence MRI images (C, D) showed that lesion at C7-T1 was dissected and removed.

Figure 2. Photographs of microscopic cross-section from the lesion at C7-T1. Histological examination of the material from the lesion at C7-T1 revealed a cyst wall line by mucin-secreting columnar epithelium (hematoxylin & eosin staining, magnification × 400).
spinal canal is an uncommon congenital lesion that usually presents with focal neurological signs [12, 15, 23-30]. Cystic changes within spinal cord pose difficult diagnostic and therapeutic problem by using computed tomography (CT) or magnetic resonance imaging (MRI). The endodermal origin of the cyst may be distinguished from the enterogenous or neuroepithelial cyst. Pathological examination reveals that the cyst wall includes the outer layer (smooth muscle and connective tissue) and the inner layer (columnar epithelium cells that produce mucin and mucopolysaccharide). It is generally considered that the increase of secretion from the cells lining the endodermal cyst cause rapid enlargement of the cyst contents, leading to spinal cord compression, therefore, an endodermal cyst should be removed as completely as possible.

The surgical approach to mass lesion in the spinal canal depends on the location and accessibility of the mass lesion. Many reports have shown that the neurenteric cyst has been operated on via a posterior approach using a laminectomy [31-34]. Despite the fact that the neurenteric cyst is usually located ventral to the spinal cord [35, 36], the study of Tuzun et al indicated that the complete cyst removal may result in damaging the spinal cord via posterior approach, and this method is generally considered to be easier and safer than the anterior approach, especially in children [31]. Takase et al reported a case of a recurrent intradural cervical neurenteric cyst operating on using an anterior approach in a 42-year-old woman [32], and she presented with a cervical neurenteric cyst that had recurred eight years after its partial removal via a posterior approach. The patient complained of pain on the lateral side of her upper arms, and a magnetic resonance imaging showed that the recurrent cyst was located ventral to the spinal cord and compressed the cord dorsally at the C4-6 level.

It is known that enterogenous cysts are uncommon congenital lesions mostly located in the lower cervical region of the spinal canal and have only rarely been observed in the upper cervical and lumbar spine region. Tuzun et al reported a case of neurenteric cyst of the upper cervical spine in an 11-year-old female child [31], and she presented with the symptoms of neck pain that radiated to both shoulders and arms. Spine MRI scans revealed a cystic mass lesion ventral to the C1-3 spinal cord without enhancement after gadolinium injection. No postoperative complications were noted after the cystic lesion was surgically removed via a posterior approach. Choi et al also reported a 15-year-old boy having an intraspinal neurenteric cyst located at cervical spine, and he presented with symptoms of neck pain and both shoulders pain for 2 months [37]. Magnetic resonance imaging of the patient showed an intradural extramedullary cystic mass at the C1-3 level without enhancement after gadolinium injection. After hemilaminectomy at the C1-3 levels was performed and the lesion was completely removed through a posterior approach, histological examination showed the cystic wall lined with ciliated pseudostratified columnar epithelium containing mucinous contents. Marchionni et al reported a case of upper cervical spinal enterogenous cyst occurred in a 76-year-old woman presenting with tetraparesis, left-sided hemisensory loss and occasional neck pain [8]. Her cervical magnetic resonance imaging showed a partially cystic lesion compressing the C2-4 spinal cord. Complete cyst resection was performed. Pathological examination revealed that of an enterogenous cyst. Arslan et al reported a 24-year-old woman with a recurrent enterogenous cyst in the L2 lumbar spinal canal [38], and she presented with lower back and left radicular leg pain for 1 year and underwent complete cyst resection via a posterior approach. She experienced an operation on for mass lesion at the same level 10 years ago. Shuangshoti et al reported a sudden paraparesis case induced by intraspinal cervicodorsal mass lesion [7]. He experienced sudden paraparesis in 6-year-old, and subsequently underwent repeated surgical explorations. At the age of 20 years, the mass lesion was subtotally removed, and the histopathological diagnosis was that the lesion was an intraspinal genuine endodermal epithelial cyst.

Conclusions

In conclusion, notwithstanding some discrepancies, complete cyst resection is generally not advocated via a posterior approach because there exists the high risk of the spinal cord injury. Further studies are needed to clarify the underlying mechanisms for the recurrence of the intraspinal enterogenous cysts after cyst aspiration and partly resection.
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Disclosure of conflict of interest

None.

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