Microsurgical Excision of the Craniocervical Neurenteric Cysts by the Far-Lateral Transcondylar Approach: Case Report and Review of the Literature

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ABSTRACT

Neurenteric cysts in the anterior craniocervical junction (CCJ) region can be found in extremely rare cases. We report one case with craniocervical neurenteric cyst that was excised by the far-lateral transcondylar (FLT) approach. A 43-year-old man presented with a history of recurrent episodes of mild neck pain and dysesthesia in his bilateral hands of 2 years' duration with rapid deterioration 3 weeks prior to admission. Magnetic resonance imaging (MRI) of the CCJ region revealed a well-defined intradural cystic lesion located ventral from the pontomedullary junction to C1 vertebra with medulla and C1 cord compression. This patient underwent total excision of the lesion via the FLT approach without any postoperative neurological deficits, and the histopathologic diagnosis was neurenteric cyst. Follow-up MRI has revealed no evidence of recurrence. The clinical features, imaging studies, and surgical approach options involved in resecting craniocervical neurenteric cysts are discussed, along with a review of the literature.

KEYWORDS: Neurenteric cyst, microsurgery, far-lateral transcondylar approach, craniocervical junction

Neurenteric cysts of the central nervous system are rare congenital lesions with epithelium-lined cyst wall, which are presumed of endodermal derivation. In the literature, neurenteric cysts generally occur in the spinal canal, making up 0.7 to 1.3% or 0.3 to 0.5% of all spinal cord tumors,^{1–3} with much lower incidences of location in the intracranial site or the craniocervical junction (CCJ) region.^{4–16} On the basis of the literature review, only 17 cases with the craniocervical neurenteric cysts treated by surgery have been reported in the

literature.^{5,7,17–29} Surgery is the usual mainstay of treatment for neurenteric cysts. With only partial resection, neurenteric cysts may recur at the original site on long-term follow-up.^{6,7,17,30,31} The incomplete resection of neurenteric cysts may be associated with postoperative malignant transformation or widespread cranial-spinal dissemination. Some authors advocate the aggressive resection to reduce the possibility of neurenteric cyst recurrence.^{32,33} The posterior approach is a common choice to attack craniocervical

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neurenteric cysts. However, because neurenteric cysts are generally located in the subarachnoid space ventral to the brain stem and spinal cord,^{1,4,14,19,22–24,30,31,34–42} the operative view of the craniocervical neurenteric cysts in this approach is usually blocked by the brain stem and cervical cord, and it is hard to perform total excision. Therefore, choosing a reasonable approach to craniocervical neurenteric cysts is the first critical step in surgical treatment. We describe our experience in the management of craniocervical neurenteric cysts excised via the far-lateral transcondylar (FLT) approach, and we review the treatment strategies discussed in the literature.

CASE REPORT

History and Examination

This 43-year-old man presented with a history of recurrent episodes of mild neck pain and dysesthesia in his bilateral hand of 2 years' duration with rapid deterioration 3 weeks prior to admission. He had no difficulty with swallowing or speech, and the strength of all extremities, gait, and coordination were normal.

The neurological examination demonstrated normal mentation and cranial nerve function, but sensory system examination revealed hypesthesia below C3 dermatome. A Babinski response was elicited bilaterally.

Neuroimaging Characteristics

Magnetic resonance imaging (MRI) of the CCJ region revealed a well-defined intradural cystic lesion in fusiform shape without any associated vertebral anomalies, located ventral from the pontomedullary junction to the C1 spinal cord. T2-weighted images displayed a hyperintense signal (Fig. 1A), and T1-weighted MRI demonstrated a hypointense mass without enhancement (Fig. 1B, C). The medulla and C1 spinal cord were found to be compressed and bulged posteriorly in the sagittal image.

Operation and Histopathologic Findings

This patient underwent surgical treatment by a right FLT approach. After completing the craniotomy with an additional C1 hemilaminectomy, a Y-shaped incision of

Figure 1 (A–C) Preoperative magnetic resonance (MR) images of the craniocervical junction region. (A) Axial T2-weighted image demonstrating a ventrally situated homogeneously hyperintense cyst. (B) Postgadolinium (post-Gd) axial T1-weighted image demonstrating a homogeneously hypointense cyst without enhancement. (C) Post-Gd sagittal T1-weighted image demonstrating neurenteric cyst at the craniocervical junction causing flattening of the brain stem and spinal cord without enhancement. (D–F) Postoperative MR images revealing total resection. (D) Axial T2-weighted image. (E) Post-Gd axial T1-weighted image. (F) Post-Gd sagittal T1-weighted image.

the dura mater was made posterior to the sigmoid sinus and vertebral artery (VA) where it pierces the dura, and extending inferiorly toward the C1 lamina. After opening the dura mater, the medulla and C1 spinal cord were exposed, flattened and bulging posteriorly. Meanwhile, most of the cyst wall was identified with thick membrane at the ventral surface of the medulla and C1 spinal cord, adhering to the 10th, 11th cranial nerve roots, which splayed over the mass. First, the lesion was gently punctured with a small needle, and the milky, viscous fluid was aspirated slowly to avoid sudden decompression of the medulla and C1 cord. After partial aspiration and decompression of the cyst, the lesion was lax and the VA adhering to the cyst wall could be easily exposed and protected. Second, the inferior aspect of the cyst was mobilized from the cranial nerve and C1 nerve roots with all the nerves left intact, and the anterior aspect was dissected off the VA. Finally, because the lesion did not involve the arachnoidal membranes of the medulla and C1 cord, we could easily separate the posterior aspect of the capsule from the lateroventral aspect of the medulla and C1 cord through gentle dissection of the well-defined plane between the lesion and normal neural tissues (Fig. 2A–F). The lesion was totally excised, and histological examination showed the cyst wall lined by a ciliated columnar epithelium with basal nuclei. Immunohistochemistry revealed periodic acid-Schiff-positive and cytokeratin-positive staining, which suggested an endodermal origin (Fig. 2G–I).

Postoperative Course

The patient's symptoms improved rapidly after surgery. This patient has been followed with serial neuroimaging, and at 3-year postoperative follow-up, clinical examination and MRI demonstrated this patient was neurologically asymptomatic with no signs of tumor recurrence (Fig. 1D–F).

DISCUSSION

Clinical and Imaging Characteristics

Neurenteric cysts most likely arise from persistent abnormal communication between endodermal and neuroectodermal tissues in the embryogenic phase, usually associated with vertebral anomalies.^{7,13,14,43} Though their sites may range from the brain stem to conus with the predilection for the lower cervical, upper thoracic, cervicothoracic junction, and thoracolumbar junction regions,^{1,4,6,14,17,20,22,30,36–38,40–42,44,45} in extremely rare cases they can be found in the anterior CCJ. Previously only 17 cases, summarized in Table 1, have occurred to the best of our knowledge.

As in the present study, most craniocervical neurenteric cysts are located ventral to the brain stem and

cervical cord, and the symptoms result from compression or inflammatory reaction by the lesions. Headache may be the most frequent complaint in the cases with craniocervical neurenteric cysts; however, there are no specific clinical features for preoperative diagnosis, and sometimes the intermittent progression of neurological symptoms may delay the diagnosis. MRI is the most effective way to explore the craniocervical neurenteric cysts; MRI can provide information on the cyst's relationship with adjacent important structures. Usually neurenteric cysts appear as low-intensity mass on T1-weighted image and high-intensity signal on T2-weighted image with no enhancement in MRI. However, these signal characteristics may vary with the high-protein content fluid within the cysts,^{43,46} and sometimes it is difficult to radiologically distinguish neurenteric cysts from other cysts such as arachnoid, dermoid, or epidermoid cysts.^{47,48}

Surgical Strategy for Craniocervical Neurenteric Cysts

Surgical excision is accepted widely as the preferred treatment for the craniocervical neurenteric cyst. The prognosis is favorable after total removal of the lesion. We also report one case of craniocervical neurenteric cyst with excellent neurological outcome after complete resection. Long-term follow-up of incompletely excised cysts shows that some cysts recur because the residual cyst wall has proliferative potential.^{6–8,14,26,29,31,49–53} Reoperation of recurrent cysts is technically difficult due to excessive scarring from the incomplete excision.^{14,23,24,29,30,54} Furthermore, Gessi et al and Perry et al have published reports on the cranial-spinal dissemination or malignant transformation of neurenteric cysts associated with incomplete resection.^{32,33} Lazareff and Hoil Parra reported that an unexcised craniovertebral neurenteric cyst may be fatal.²⁴ Hence, most neurosurgeons advise that the first choice of surgical treatment for neurenteric cyst is the early complete resection with preservation of functional neural tissue of the brain stem and spinal cord.^{13,14,20,24,31–33,38,55}

We also agree that the major determinant of the degree of craniocervical neurenteric cyst resection is the presence of a clear plane between neurenteric cysts and the brain stem, spinal cord, nerve, and critical vessels. Most craniocervical neurenteric cysts are oval or fusiform in shape, with a well-defined plane identified along the cyst interface with the brain stem and cervical cord. However, for the cases with cyst wall strongly adhering to the surrounding neurovascular structures, the adhesion may preclude complete resection of the cysts. In these cases, there is no need to pursue the total resection, considering the risk of injury to the functional neural tissue.^{20,26,36,42,56,57} Some authors have recommended a cystosubarachnoid shunt

Figure 2 (A–F) Intraoperative photographs of the neurenteric cyst (NC) in the far-lateral transcondylar (FLT) approach. (A) Intraoperative photograph shows retroauricular curvilinear skin incision of the FLT approach. (B) A well-encapsulated lesion is exposed, adhering to the 10th, 11th cranial nerves, and the medulla and cervical cord are found to be flattened and bulging posteriorly. The nerve roots are displaced superiorly and splayed over the mass. (C) The lesion is punctured and the milky, viscous fluid is aspirated. (D) The cyst is lax and the vertebral artery (VA) is exposed after partial aspiration and decompression of the cyst. (E) The cyst wall is dissected from the medulla. (F) The cyst is excised totally with the medulla, cervical cord, VA, cranial nerve, and C1 nerve intact. (G–I) Photomicrography of the NC wall appearance. (G) Histological examination shows the cyst wall lined by ciliated columnar epithelium with basal nuclei (hematoxylin and eosin, $\times 200$). (H) Immunohistochemistry reveals a strong positive reaction for cytokeratin, suggesting the epithelial nature of these cells (keratin staining, $\times 200$). (I) The epithelium contains mucin-secreting goblet cells with material in the supranuclear cytoplasm staining positive with periodic acid-Schiff (periodic acid-Schiff, $\times 200$). *, NC; ★, the medulla; ☆, the C1 cord; △, the vertebral artery; ▲, the dura mater ventral to the medulla after NC total resection.

with partial cyst wall resection to prevent the possibility of severe neurological deficits.²⁹ All patients with a small residual remnant of the cyst wall, which may be responsible for recurrence, require long-term clinical follow-up with serial MRI to detect the recurrence as early as possible.

Surgical Approach Options to Craniocervical Neurenteric Cysts

Although complete excision of the craniocervical neurenteric cysts should be attempted to prevent recurrence during the primary operation, it is not always possible because the cysts are situated ventral to the brain stem

Table 1 Cases of the Craniocervical Neurenteric Cysts in the Literature

First Author and Year	Age/Sex	Symptoms	Cyst Position	Surgical Approach	Type of Resection	Recurrence
Abe 1999 ¹⁷	60/F	Headache and limitation of neck movement	CCJ	Transcondylar approach	Total	No
Breeze 1990 ⁵	37/M	Neck pain	CCJ	Occipital approach with C1 laminectomy	Partial	No
de Oliveira 2005 ⁷	1/M	Meningitis	CCJ	Post-fossa approach (1st) Transoral approach (2nd)	Partial (initial) Total (reoperation)	Yes No
Ergün 2000 ¹⁹	3/M	Weak in four extremities and neck pain	From CCJ to T1	C3–C4 laminectomy	Partial	No
Filho 2001 ²⁰	30/F	Headache and nausea	CCJ	Suboccipital retrosigmoid and C1 hemilaminectomy	Total	No
	30/F	Seizure	CCJ	Suboccipital craniectomy and C1 hemilaminectomy	Subtotal	No
	25/M	Headache	CCJ	Suboccipital retrosigmoid and C1 hemilaminectomy	Subtotal	No tumor grow
Fuse 1998 ²¹	50/M	Headache and neck pain	CCJ	Transoral approach	Total	No
	49/M	Headache	CCJ	Suboccipital approach with C1 laminectomy	Total	No
Harris 1991 ²²	30/F	Headache, nausea, and blurred vision	CCJ	Retromastoid approach	Total	Unknown
Koksel 1990 ²³	40/M	Neck pain and weakness	CCJ	Transoral approach	Total	No
Lazareff 1995 ²⁴	7/F	Neck pain	CCJ and cervical cord	C2–C3 laminectomy	Total	No
Lin 2004 ²⁵	45/F	Headache and diplopia	CCJ	Subtemporal approach	Subtotal	Unknown
Liu 2005 ²⁶	46/F	Pain in right shoulder, right ear	CCJ	Transcondylar approach	Total	No
	26/F	Headache	CCJ	Transcondylar approach	Total	No
Rao 1996 ²⁷	10/F	Neck pain	CCJ and cervical cord	C5–C6 hemilaminectomy	Partial	Unknown
Sakata 2008 ²⁸	1/M	Weak in four extremities	CCJ	Suboccipital retrosigmoid and C1 hemilaminectomy	Total	No

CCJ, craniocervical junction; F, female; M, male.

and cervical cord, blocking the operative view. Hence, choosing a reasonable approach to the craniocervical neurenteric cyst is the first critical step in the surgical treatment. Many different surgical approaches to gain access to neurenteric cysts in the anterior CCJ have been discussed in the literature. Most cases are excised by a conservative posterior approach, such as the suboccipital approach, despite the fact that the craniocervical neurenteric cyst is situated ventral to the brain stem and cervical cord.^{5,7,19,20,24,27,28} As part of the cyst wall adheres to the ventral cord, it is difficult to visualize the cyst posteriorly and resect it totally. Even though at times the ventrally situated cysts can be approached posteriorly, total excision is sometimes associated with high risk to the brain stem or cord damage, especially when manipulating and retracting the medulla or spinal

cord. Several other authors have advocated an anterior approach, such as the transoral approach, for the craniocervical neurenteric cyst total resection.^{21,23,30} The transoral approach can access to the lesion in the anterior CCJ with good visualization; however, it is technically difficult and is usually associated with longer operation time and higher risk of postoperative complication.^{36,53} Menezes and Ryken have also reported an unsuccessful case resected by the transoral approach, which was reoperated by the dorsolateral approach.³⁰

The FLT approach has proved to be an effective approach to manage the lesion involving the anterior CCJ region, providing an unobstructed view of the lateroventral cervicomedullary area.^{26,58,59} On the basis of a literature review, only three excisions by the FLT approach of neurenteric cysts in the CCJ region have

been previously published.^{17,26} However, we prefer the FLT approach for total excision of the craniocervical neurenteric cysts based on the experience of the successful case described here. This approach can provide a direct operative view of the lower clivus and anterior craniovertebral junction, and neurenteric cysts at the ventral surface of the brain stem and cervical cord can be easily attacked in the operation. Furthermore, without retracting the medulla and cervical cord, we can assess the ventrally and ventrolaterally located cyst attachments and separate them directly in the FLT approach. Though the optimal surgical approaches to the craniocervical neurenteric cysts remain controversial, we believe that the FLT approach is indicated for the following reasons: better operative view of neurenteric cysts in the ventral dural space of the lower clivus and foramen magnum, and less risk of trauma to the brain stem and cervical cord.

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