## **Observable Recurrence of Cervicothoracic Neurenteric Cyst after Subtotal Resection: A Case Report**

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Neurenteric cysts (NECs) are rare, congenital malformations of the central nervous system that are lined by an epithelium of an intestinal nature. They are benign lesions often found in the cervical and upper thoraco-cervical areas<sup>1)</sup>. The lesions commonly present in the second decade of life and are usually located ventral or ventrolateral to the cord in the intradural compartment. According to a recent review of postsurgical outcome analyses, incomplete posterior surgical excision carries a higher cumulative risk of recurrence as the follow-up period increases<sup>2)</sup>. However, some previous reports described cyst recurrences that needed revision surgeries<sup>3,4)</sup>. Asymptomatic cases with cyst recurrence are not well-known. We report here cervical/thoracic intradural extramedullary NECs that were surgically removed and recurred without neurological symptoms.

A 4-year-old girl presented with neck pain that had persisted for 1 week. She complained of severe neck pain with difficulty urinating at the time of admission to our hospital. Neurological examination revealed spastic paralysis in both lower extremities without sensory disturbance. Magnetic resonance imaging (MRI) revealed an intradural cyst located anterior to the spinal cord at the C3 to C5 level (Fig. 1A-C). A hemilaminectomy was performed at C2 to C6. The spinal cord shifted posteriorly. A gray-white cyst appeared anterior to the spinal cord. After the cyst wall was punctured, milky white fluid was aspirated. After the operation, there was immediate improvement in the neck pain and urinary disturbance. However, 2 months later, she complained of severe neck pain again with muscle weakness in the right upper extremities. MRI revealed cyst recurrence (Fig. 1D, E). During the second surgery, we detached the cyst wall from the spinal cord, following excision. We tried to completely remove the cyst wall but found severe adhesion between the cyst wall and the pia mater. Therefore, complete removal of the wall would have involved spinal damage. Thus, we performed a cyst wall resection (Fig. 2A). Histopathological examination of the tissue demonstrated a cyst wall lined with cuboidal to columnar epithelium, suggesting a type A Wilkins and Odom neurenteric cyst histopathological classification<sup>5</sup> (Fig. 2B). The patient subsequently recovered from the neurological deficit. The cyst recurred 1 year later. We monitored her for 11 years without any symptoms, and the recurrent cyst has diminished in size year by year. (Fig. 1F-H).

Here, we presented a NECs case that underwent surgical treatment. Although gross removal of the cyst wall was achieved, the cyst recurred without symptoms after the surgeries. During postoperative follow-up, no neurological symptom was observed. Although we performed reoperation through the previous laminectomy, it would be better to carry out laminoplasty to get a wider surgical field, enabling us to remove the cyst as much as possible<sup>6</sup>.

Surgical treatment in NECs should be done to prevent spinal irritation. Indeed, the dorsal wall of the cyst was adhered tightly to the spinal cord in our case. Miyoshi et al reported that if the adhesion was tough and some portion of the cyst was intramedullary, complete resection could result in postoperative neurologic deterioration<sup>7</sup>. Moreover, several previous reports advised against overaggressive removal of the lesion because of the benign course after subtotal resec-

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**Figure 1.** Case 1, 4-year-old girl. MRI showing the intradural extramedullary cystic mass lesion at C3/5. The spinal cord was completely stretched out over this ventral cyst. (A) T1-weighted sagittal image. (B) T2-weighted es agittal image. (C) T2-weighted axial image. Postoperative MRI showing cyst recurrence. (D) T2-weighted sagittal image. (E) T2-weighted image. After revision surgery, the cyst recurred (arrow). (F) T2-weighted sagittal images 1-year postsurgery. (G) Seven years postsurgery. (H) Eleven years postsurgery.

tion <sup>4,8,9)</sup>. Therefore, we could hypothesize that subtotal cyst wall resection could be achieved with a good prognosis as the lesion is benign. In conclusion, early surgical intervention and long-term follow-up after partial excision are recommended, particularly to prevent long-term morbidity. If the cyst wall is adhesive to the pia mater, partial resection would be desirable to prevent spinal damage. Partial resections would indicate good prognoses.

**Conflicts of Interest:** The authors declare that there are no relevant conflicts of interest.

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**Figure 2.** (A) Intraoperative microscopic view shows severe adhesion between the cyst wall and pia mater. (B) Histopathological examination showing cystic wall as cuboidal to columnar epithelial cells.

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## References

- **1.** Devkota UP, Lam JMK, Ng HK, et al. An anterior intra-dural neurenteric cyst of the cervical spine: complete excision through central corpectomy approach: case report. Neurosurgery. 1994;35 (6):1150-4.
- 2. Kida K, Tani T, Kawazoe T, et al. Case report a recurrent cervical neurenteric cyst treated anteriorly: safe, gross-total excision facilitated by prophylactic unilateral vertebral artery exposure, microdissection, and spinal cord monitoring—A case report and technical note. Case Rep Orthop. 2018;2018:7620182.
- **3.** Holmes GL, Trader S, Ignatiadis P, et al. Intraspinal enterogenous cysts. A case report and review of pediatric in the literature. Am J Dis Child. 1978;132(9):906-8.
- **4.** Kim CY, Wang KC, Choe G, et al. Neurenteric cyst: Its various presentations. Childs Nerv Syst. 1999;15(6-7):333-41.
- **5.** Lippman CR, Arginteanu M, Purohit D, et al. Intramedullary neurenteric cysts of the spine: case re- port and review of the literature. J Neurosurg. 2001;94(2):305-9.
- **6.** Savage J, Casey J, McNeill I, et al. Neurenteric cysts of the spine. J Craniovertebr Junction Spine. 2010;1(1):58.
- **7.** Miyoshi K, Nakamura K, Hoshino Y, et al. Removal of enterogenous cyst of the cervical spine through anterior approach. J Spinal Disord. 1988;11:84-8.
- **8.** Takahashi S, Morikawa S, Saruhashi Y, et al. Percutaneous transthoracic fenestration of an intramedullary neurenteric cyst in the thoracic spine with intraoperative magnetic resonance image navigation and thoracoscopy. J Neurosurg Spine. 2008;9:488-92.
- **9.** Takase T, Ishikawa M, Nishi S, et al. A recurrent intradural cervical neurenteric cyst operated on using an anterior approach: A case report. Surg Neurol. 2003;59:34-9.

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