



Cervical Intradural Enterogenous Cyst Containing Nevus Cells: A Case Report

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Abstract

Introduction: Enterogenous cysts are rare developmental malformations located predominantly in the spinal canal. We report here a very rare case with enterogenous cyst, containing nevus cells pathologically.

Case Presentation: This was a case of a 48-year-old man complaining of gradually developing numbness in the right upper and lower limbs for two months. Magnetic resonance imaging revealed a noncontrast-enhancing mass lesion at the C2-C4 level, that are low signal intensity on T1-weighted imaging, high signal intensity on T2-weighted imaging. Computed tomography myelogram showed a positive meniscus sign with no contrast enhancement. We performed subtotal resection of the cyst *via* posterior approach. Histopathology showed mucin-producing simple columnar or cuboidal ciliated cells, which was consistent with an enterogenous cyst. Interestingly, nevus cells were observed in the stroma surrounding the cyst epithelium. The patient's numbness and hypoesthesia improved immediately, and had no recurrence for two years.

Discussion: This is the third case describing enterogenous cyst, containing nevus cells, and these findings suggested enterogenous cysts are endodermal in origin.

Conclusion: We report a very rare case of an enterogenous cyst containing nevus cells pathologically. Our findings suggest that enterogenous cysts are endodermal in origin.

Keywords: Spine, Tumour, Neural tube defects, Neuroenteric cyst, Endoderm, Nevus

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Introduction

Enterogenous cysts are rare developmental malformations located predominantly in the spinal canal. These cysts are typically lined by columnar epithelium resembling enteric epithelium or columnar ciliated epithelium resembling respiratory epithelium [1]. As far as we know, there have been only two cases reported, containing melanocytes in the stroma surrounding the enterogenous cyst [2]. We report here a very rare case of an enterogenous cyst, containing nevus cells pathologically.

Case Presentation

This was a case of a 48-year-old man complaining of gradually developing numbness in the right upper and lower limbs. He has no concerns with relevant details of related past interventions. His symptoms had revealed two months before without any apparent reason. After Magnetic Resonance Imaging (MRI) showed an intradural and extramedullary mass lesion in the cervical spine, he was referred to our institution. Neurological examinations revealed slight hypoesthesia in the right fingers and foot, but there was no muscle weakness in any of the extremities. Dysfunction of the bladder and bowel was absent. Deep tendon reflexes were normal. On sagittal MRI, the mass lesion showed low signal intensity at the level of C2-C4 on T1-weighted imaging, high signal intensity on T2-weighted imaging, and high signal intensity on short Fluid-Attenuated Inversion Recovery (FLAIR) imaging. Gadolinium-enhanced MRI revealed no change. The mass was approximately 3.5 cm × 2.5 cm in size and was located in the subarachnoid space at the left side anterior to the spinal cord (Figure 1). Computed Tomography Myelogram (CTM) showed a positive meniscus sign with no contrast enhancement (Figure 2). From these myelographic (CTM and MRI) findings, an intradural and extramedullary cystic lesion was suspected. Other diagnosis we considered was arachnoid cyst and perineural cyst, but they should be enhanced by CTM. We planned the resection of the cyst *via* posterior approach, using transcranial Motor Evoked Potentials (MEPS), for the intraoperative spinal cord.

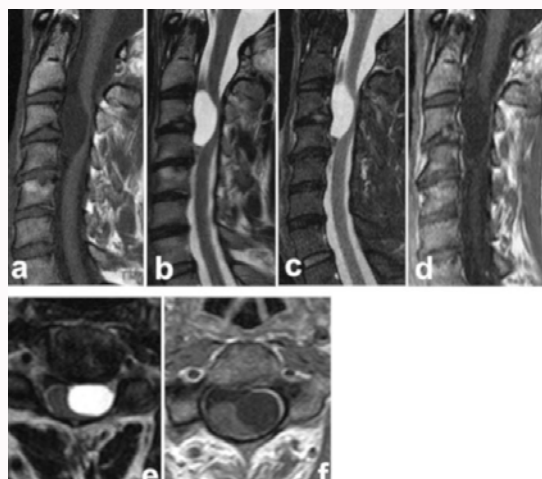


Figure 1: Preoperative MRI. **a:** Sagittal T1-weighted image showing a low intensity area at C2-C4 level. **b:** Sagittal T2-weighted image showing a high intensity area at C2-C4 level. **c:** Sagittal short Fluid-Attenuated Inversion Recovery (FLAIR) image showing a high intensity area at C2-C4 level. **d:** Sagittal gadolinium-enhanced T1-weighted image showing a low intensity area at C2-C4 level. **e:** Axial T2-weighted image showing an intradural mass lesion located in the subarachnoid space at the left side anterior to the spinal cord. **f:** Axial gadolinium-enhanced T1-weighted image.

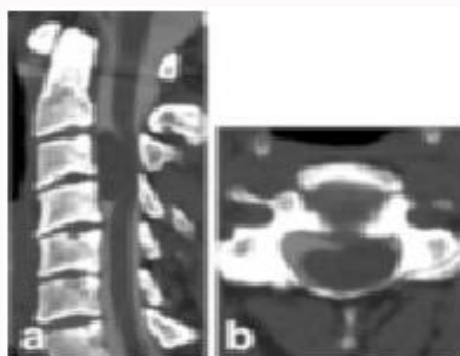


Figure 2: CTM indicated an intradural extramedullary mass, showing no contrast enhancement. **a:** Sagittal, **b:** Axial.

After dissection of the left-side paravertebral muscles, hemilaminectomy of C2-C4 was performed. A reddish-brown cystic mass was found when the dura was opened along the left side from the middle. The cystic mass adhered slightly to the ventral spinal cord and dura, and careful detachment was needed (Figure 3). The fluid content of the cyst was first evacuated through needle aspiration; after which, the collapsed walls were removed as completely as possible. The cyst fluid was clear and colorless mucinous in consistency.

Histopathological presentation of the Hematoxylin Eosin (HE) stained specimen's revealed aggregated mucin-producing simple columnar or cuboidal ciliated goblet cells surrounding a central cystic cavity, which was consistent with an enterogenous cyst. Nevus cells were observed in the stroma surrounding the cyst epithelium, which was immunohistochemically stained with Melan-A (Figure 4).

The postoperative clinical course was well, and numbness and hypoesthesia improved immediately. The patient has been doing well and had no recurrence for two years after surgery.

Discussion

The term enterogenous cyst was used for the first time by

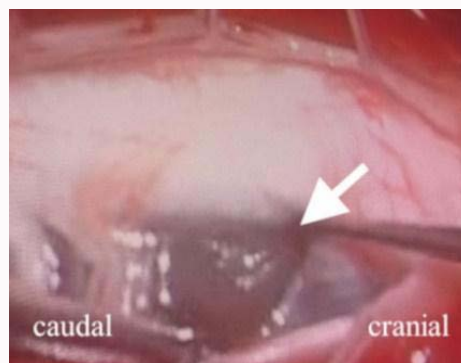


Figure 3: Intraoperative findings. The cyst (white arrow) was reddish-brown and adhered slightly to the ventral spinal cord and dura.

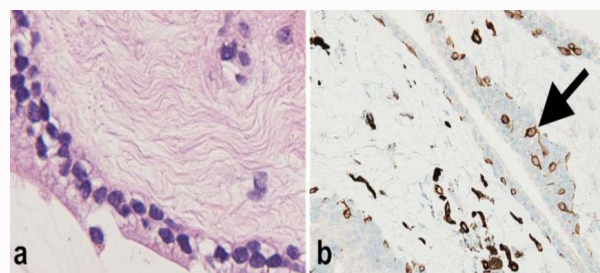


Figure 4: Histopathological findings. **a:** HE-staining: Mucin-producing simple columnar or cuboidal ciliated goblet cells surrounding a central cystic cavity (original magnification, x400). **b:** Immunohistochemical staining with Melan-A. Melanocytes were present in the stroma surrounding the cyst (original magnification, x400).

Harriman to describe lesions previously known as neuroenteric, endodermal, or respiratory cysts [1]. Enterogenous cysts are rare lesions, comprising 0.7% to 1.3% of spinal axis tumors [3]. The general location of these cysts is the subarachnoid space anterior to the spinal cord. More than 90% of the reported cases are located in the intradural and extramedullary compartment, whereas less than 5% of lesions are found within the intramedullary compartment [3,4].

Total resection is the ideal surgical treatment for extramedullary intraspinal cysts to prevent cyst recurrence [5]. Postsurgical recurrence rate has been reported to range from 0% to 37%. Kim et al. and Cai et al. reported no recurrence in their series of eight and seven cases, respectively, whereas Chavda et al. observed a 37% recurrence among eight patients [6-8]. It is noteworthy that recurrence was observed in cases in which complete resection of the cysts was impossible because of extensive adhesion to neural anatomy or spinal anomalies [8]. Among various surgical approaches reported (anterior, posterior, and lateral), posterior resection is the most frequently used method. Although enterogenous cysts usually present anterior to the spinal cord, which will obscure the cyst structure, the posterior approach will produce good access to the lesions by aspiration of the cyst content and cord manipulation with fewer complications [9,10]. In our case, in which the cyst was localized in the ventral position at C2-C4 level, we employed posterior approach and performed subtotal resection. As the adhesion to neuronal tissue was mild and limited, we were able to cut off the cyst wall almost completely after aspiration of the fluid content. Careful manipulation of the cord, using intracranial MEPS, was needed to avoid the risk of spinal cord injury. The patient has no neural complications or recurrence two years after surgery.

Enterogenous cysts of the central nervous system are lesions of

controversial pathogenesis or embryological origin [8,11]. Their association with spinal cord or vertebral abnormalities has been cited as evidence of the embryonic endodermal origin [12]. Others have suspected teratomatous origin, citing the midline location [13]. In our case, ciliated and mucinous epithelia, both of endodermal origin, were observed in enterogenous cysts. Presence of mucin-producing epithelium would support an endodermal, rather than neuroectodermal, origin [14].

Nevus cells were observed in our case within the fibrous stroma surrounding the cyst epithelium. As far as we know, there have been only two cases reported containing melanocytes in the stroma surrounding the enterogenous cyst [2]. Melanocytes normally exist in the leptomeninges, and this finding suggests that the stroma tissue is arachnoidal in origin, not a mesodermal component of a teratomatous lesion.

Conclusion

We report a very rare case of an enterogenous cyst containing nevus cells pathologically. Our findings suggest that enterogenous cysts are endodermal in origin, rather than the neuroectodermal or mesodermal components characteristic of teratomatous lesions.

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