

Heterotopic neuroglial tissue in lumbar spinal canal – Case report

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Abstract

Ectopic and heterotopic both refer to cells displaced from their normal site – ectopic cells occur outside their organ of origin whereas heterotopic cells are in an aberrant location within their organ of origin. The most common localization of heterotopic neuroglial tissue (HNT) is the nasal region. We report a case of 69-years-old man who presented with long history of lumbosacral pain with paresis of the inferior limb. MRI examination detected a cystic tumour located at the L1-L2 level. Histologic examination revealed the presence of heterotopic neuroglial tissue, containing synaptophysin, glial cells and encircled meninges.

Key words

heterotopic neuroglial tissue, cystic tumour, lumbar spine canal

INTRODUCTION

Ectopic and heterotopic both refer to cells displaced from their normal site – ectopic cells occur outside their organ of origin whereas heterotopic cells are in an aberrant location within their organ of origin [1]. Neuroglial heterotopia, ectopic brain tissue, or differentiated neural tissue outside the physiological localization is uncommon, and these anomalies most commonly occur in the nasal region. However, ectopic brain tissue has been reported to occur in the pharynx, lung, orbits, palate, tongue, cheek, lip and neck [2,3]. To the best of our knowledge, no incidence of heterotopic brain tissue adjacent to the spinal cord in the lumbar region has been reported. The following case is all the more noteworthy because the abnormal tissue led to paresis of the inferior limb. MRI examination detected a cystic tumour located at the L1-L2 level.

CASE REPORT

History and examination. The patient was 69-year-old man who presented with a long history of pain in the lumbosacral region. For about a month and a half, the patient had been experiencing increasing back pain with radiation to his right leg. Patient had a congenital lumbosacral spinal deformity and skin defect located in lumbosacral region of spine, pilonidal cyst. Neurological examination showed decreased mobility and strength in the right inferior limb, IV/V grade in Lovett scale, with atrophy of all muscle groups within the limb. Additionally, the patient reported hypoesthesia of the buttock and superior region of the thigh of right leg, and bowel incontinence,

Imaging findings. An MRI study of the lumbar spine demonstrated cystic lesion measuring 2.5 x 0.7 x 0.9 cm,

located at the cauda equina at the L1–2 level. The lesion was well circumscribed, hypointense in comparison to the spinal cord on T1-weighted images, with gadolinium enhancement and hyperintense on T2-weighted images (Fig. 1).

Surgery and postoperative course. An L1 and L2 laminectomy was performed and the ligamentum flavum was excised. Dura mater was tense over the bulge of the tumour. Dura was opened with a midline incision. The lesion within the capsule contained a thick, whitish fluid of cream-like consistency. Fluid of the cystic tumour was aspirated and the capsule excised. Neither destruction of bone nor any abnormality in the conus medullaris and nerve roots of the cauda equina were observed. The patient awoke with the same overall and neurological status as the before surgery. He was discharged in good condition after 5 days of postoperative hospitalization. On the month postoperative follow-up, patient reported reduction of back pain and hypoesthesia.

Pathological findings. Tissue of the cystic lesion obtained at surgery was fixed in 10% buffered formalin (pH=7.4) and processed routinely through dehydration with graded alcohol, acetone, xylene and then embedded in paraffin blocks. Four-micron-thick sections were stained with haematoxylin and eosin (H&E), followed by other histochemical procedures, such as Van Gieson and PAS. Sections for immunohistochemistry were mounted on slides coated with organosilane (Dako Silanized slides, Code No. S3003) to prevent floating of specimen during immunohistochemical procedure. These sections were stained immunohistochemically using EnVision™ Detection System (Dako Real™ EnVision™ Detection System, Peroxidase/DAB+, Rabbit/Mouse, Code No. K5007) with the following Dako antibodies: S-100 (Code No. Z0311; Dilution 1:100), GFAP (M0761; 1:100), Synaptophysin (M0776; 1:20), EMA (M0613; 1:50), MIB-1 (M7240; 1:150) and CD34 (M7165; 1:50).

The surgical specimens were grey in colour, fragmented, and 0.5 cm in total diameter. Postoperative histopathological examination revealed the presence of mature synaptophysin-

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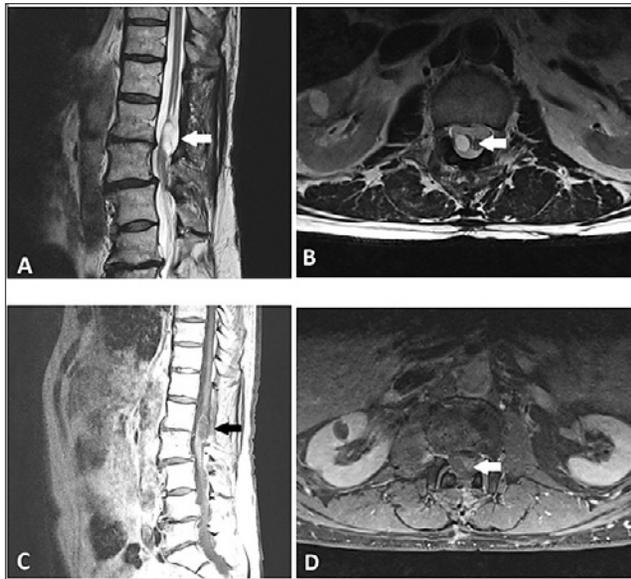


Figure 1. ??

positive neurons in the centre of the lesion (Fig. 2 C,I) and GFAP-positive glial cells on the periphery (Fig. 2 A,H). Both components were also S-100-positive (Fig. 2 G). Only a few of glial cells were immunopositive in reaction with

proliferative marker MIB-1. On the surface of the lesion there were meninges with a layer of meningotheial cells (Fig. 2 A), which focally formed clusters of cells (Fig. 2 B). Additionally, there were small and dispersed amyloid bodies (Fig. 2 D,E). A small cyst lined with meningotheial cells was visible within the largest piece of the specimen (Fig. 2 F).

DISCUSSION

Heterotopic neuroglial tissue was first described by Reid in 1852 [4]. The first case of glial heterotopia over the dorsal surface of the cervical spinal cord was recorded in 1907 by Wolbach [5]. The pathogenesis of this kind of tissue is unclear. Several mechanisms have been proposed and it may derive from encephalocele [4]. This may be due to separation of extracranial embryonic neuronal tissue through changes during cranial closure, or it may derive from the isolated remains of pluripotent neuroectodermal cells that created mature neuronal tissue. HNT is the most frequent localisation, when diagnosed in infants; however, it may also be rarely be found in adults [5,6]. Neuroglial heterotopia, ectopic brain tissue, or differentiated neural tissue most commonly occur in the nasal region. However, heterotopic brain tissue has been reported to occur in the pharynx, lung, orbits, palate, tongue, cheek, lip and neck [2, 3].

A. Conner et al. have described a 29-year-old woman

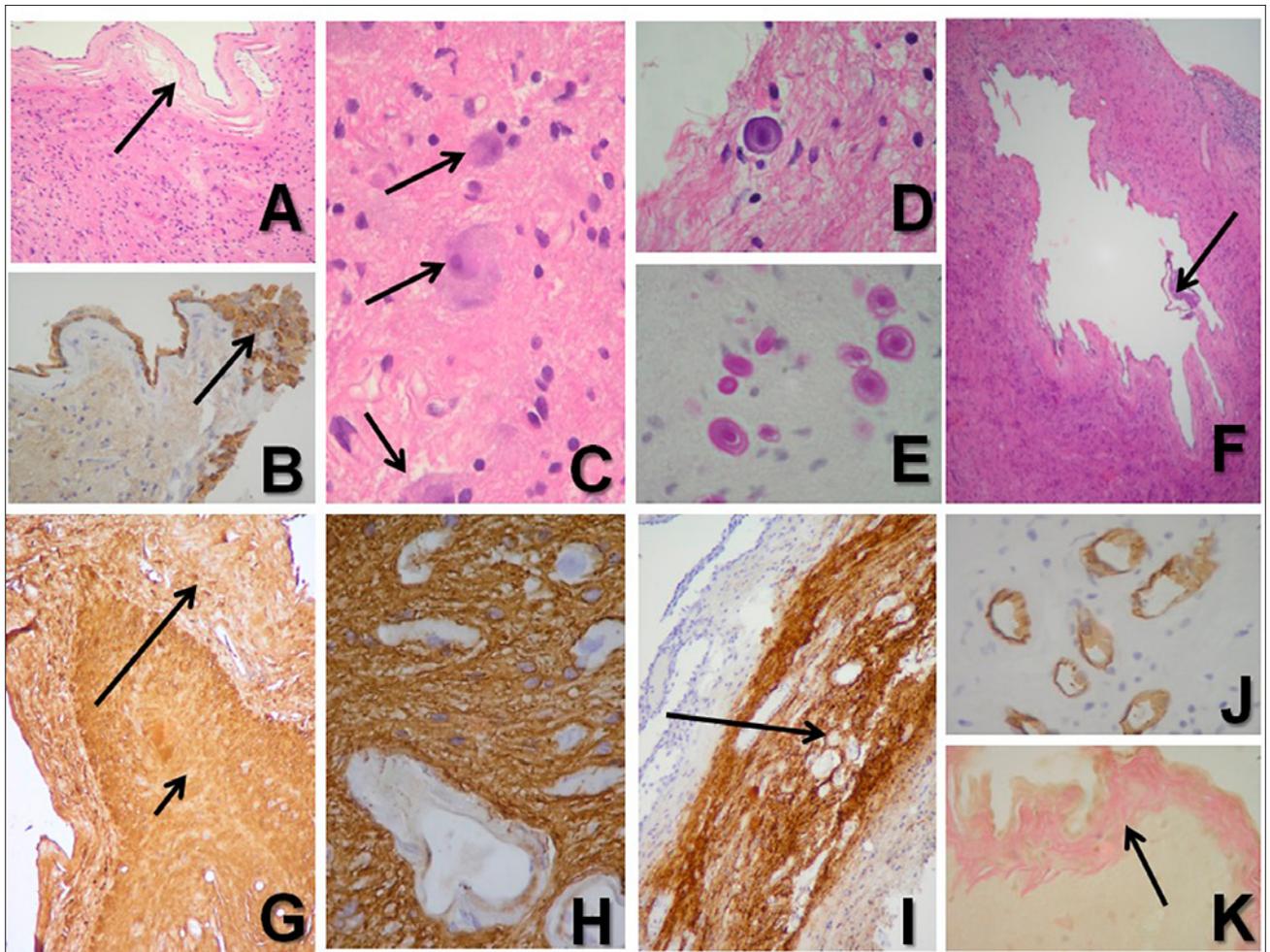


Figure 2. ??

with new-onset pain and numbness in her right leg, and urinary incontinence, in whom imaging demonstrated an enhancing mass located at the L1–2 interspace. Histological section showed an ectopic ganglion that included ganglionic neurons rimmed by sustentacular cells and embedded in a background of Schwann cells [7]. B. Shrestha et al. have described large mature brain tissue in the sacrococcygeal spine in a newborn baby girl [8].

CONCLUSIONS

In summary, heterotopic neuroglial tissue in the lumbar spine canal is a very uncommon lesion, and pre-operative diagnosis of this lesion is difficult. Imaging studies and histologic examination are the necessary investigations for diagnosis. Surgical treatment is the best choice for a symptomatic patient with pathological lesion in the spine canal. The outcome is similar to other benign lesions in this location.

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