Lumbar Intradural Extramedullary Bronchiogenic Cyst

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Key Words

Bronchiogenic cyst, surgery · Intraspinal · Pathology

Abstract

The pathological findings of an intradural and extramedullary cyst at the L1 level of the spinal canal are described in a 28-year-old male who presented with chronic lumbago and progressive weakness and numbness in both lower limbs. Histopathological examination revealed the diagnosis of bronchiogenic cyst. Bronchiogenic cysts in the spinal canal are uncommon, and cysts at the L1 level are extremely rare. Their pathogenesis is still poorly understood. They are thought to be a malformation arising from a split notochordal syndrome. We conclude that intradural extramedullary bronchiogenic cysts may appear also at lumbar levels. Surgical resection can be achieved with good outcome.

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Case Report

A 28-year-old male presented with chronic lumbago for a year and progressive weakness and numbness in both lower limbs for 2 weeks. There were no urinary or defecation problems.

The neurological examination showed hypoesthesia and 4/5 muscle strength of his right lower extremity. The bilateral tendon reflexes of the lower extremity were overactive. Babinski could not be elicited on either side. Magnetic resonance imaging of the lumbar vertebrae revealed an intradural extramedullary mass located at the L1 level (fig. 1).

Fig. 1. The arrow shows an intradural extramedullary cyst at the L1 level.

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0014-312X/08/0401-0026\$24.50/0

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A laminectomy of L1 and partial T12, L2 was performed. The lesion was well exposed by a longitudinal opening of the dura along its midline. It was a fluid-containing, $2.0 \times 1.7 \times 1.5$ cm cystic lesion, which was adherent to the dorsal conus medullaris and the cauda equina. Yellow viscous liquid in the cyst was sucked out. The cystic wall was partially resected and was sent for pathological examination. Total resection was impossible due to the inevitable injury to the conus and cauda equina. No postoperative complications occurred. The preoperative symptoms disappeared completely.

Pathological Findings

The specimen was a unilocular, thin-walled cyst, the inner surface of which was lined with pseudostratified ciliated columnar epithelium. Within the subepithelial stroma mucous and serous glands were present, which were found regularly in the normal tracheobronchial tract (fig. 2–4).

Discussion

The intraspinal cysts have various types. The here described cyst has the histopathological appearance with the ciliated lining epithelium and the wall containing mucous and serous glands. It was referred to as bronchogenic cyst [1], similar to that of the mediastinum [2, 3]. Ho and Tiel [4] further confirmed the bronchial characteristic of such a cyst by ultrastructural research. In their study 6 cell types were identified, namely ciliated cells, nonciliated cells, goblet cells, basal cells, Kulchisky's cells and undifferentiated cells. The cell types and their topographic distribution are similar to that of the human bronchial epithelium. They also considered that the epithelium of the bronchiogenic cyst is consistent with the upper respiratory tract, particularly the tracheobronchial portion, because of the absence of Clara cells, which in humans are found only in the bronchioles.

Bronchiogenic cysts are developmental abnormalities of the central nervous system [5]. The definite developmental mechanisms of such cysts are still unknown. However, they are generally believed to derive from an

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Fig. 2–4. The serous gland (arrow 2) and mucous gland (arrow 1) are well shown. Pseudostratified ciliated columnar epithelium is shown by arrow 3. **Fig. 2** $\times 100$. **Fig. 3, 4** $\times 400$.

anomalous embryological connection between the primitive foregut and the developing neural tube, probably in association with splitting or reduplication of the intervening notochord [6].

Bronchiogenic cysts have been found attached to the sternum, skin, stomach, pericardium and mediastinum, whereas intraspinal cases are extremely uncommon. Baumann et al. [7] reported an intraspinal bronchiogenic cyst and conducted a literature review in 2005. In all the cases in his review, the lesion localized at a cervical or upper thoracic level except 1 at the T12–L1 level reported by himself. The here presented cyst at the L1 level was exceedingly rare.

Bronchiogenic cysts usually grow slowly due to the tight junctions between the epithelial cells which may

limit the amount of expansion [8]. There are no instances of canceration developing within these intraspinal bronchiogenic cysts, but malignancies of the cyst in the mediastinum, including adenocarcinoma and rhabdomyosarcoma, have been reported [9, 10]. The most effective management to intraspinal bronchiogenic cysts was surgical resection. The patient's symptoms usually relieve, as we reported, after the cyst is removed. Total resection of the cyst is the goal of surgery. Nevertheless, this has not been achieved in most of the cases. Only 36% of the cysts reported in the literature have been removed totally [11], either because of the ventral location related to cord or because of the adherence of a part of the cyst to the medullary substance.

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