CASE REPORT

Intradural Intramedullary Bronchogenic Cyst (Neurentric Cyst) in Dorsal Spine

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ABSTRACT

Bronchogenic cysts are considered the 2nd most common foregut duplication cyst after neurenteric cysts and the most common non-neoplastic mediastinal cysts. These are congenital malformations derived from anomalous budding of the embryonic foregut. We report a case of 55 year old male presented with complaints of neck pain and weakness in both lower limbs. MRI revealed heterogenous enhancing SOL at D1-2veretebral level suggestive of a glial tumor (?Astrocytoma) with diffuse disc bulging causing indentation of thecal sac, spinal cord and lateral recess at C5-6 and C6-7.Laminectomy and tumor decompression was done. The resected tissue on histology showed a bronchogenic cyst. To best of our knowledge it is third case of intradural and intramedullary bronchogenic cyst seen at dorsal spine.

Key words: Intramedullary, Bronchogenic, cyst

INTRODUCTION

Bronchogenic cysts are congenital malformations derived from anomalous budding of the embryonic foregut. It is considered the 2nd most common foregut duplication cyst after neurenteric cysts.[1]These are also considered as the most common non-neoplastic mediastinal cysts.[2] These are usually solitary, but multiple may be seen and can be filled with fluid or proteinaceous material. These have been also reported in more remote locations like the neck, abdomen and retroperitoneal space. Intraspinal bronchogenic cysts are extremely rare and most of them are extramedullary.[3,4]Most of them are located in the cervical or upper thoracic region.

We present a unique case of intradural intramedullary intraspinal bronchogenic cyst at C7-D2 level. To the best of our knowledge, so far only two cases of intramedullary intraspinal bronchogenic cyst reported.[5,6]We therefore present third case of intradural intramedullary spinal bronchogenic cyst.

CASE REPORT

A 55 year old male presented with complaints of neck pain, burning sensation and numbness in both lower limbs from past six months.On clinical examination there was weakness in both lower limbs with power of 3/5. Deep tendon reflexes were brisk in both lower limbs. No sensory deficit observed. No sign and symptoms of bladder and bowel involvement were observed. There was no history of fever, tuberculosis or any previous surgery.

Magnetic resonance imaging (MRI) of patient's spine showed area of cavitation in C5-7 vertebral level-syrinx. Heterogenous enhancing SOL at D1-2veretebral level with diffuse disc bulging causing indentation of thecal sac, spinal cord and lateral recess at C5-6, C6-7 suspecting it to be a glial tumor ?Astrocytoma.

C7-D2 laminectomy and tumor decompression was done and resected tissue was sent for histopathological examination.Histopathological examination of the specimen with hematoxylin and eosin staining showed the cyst wall lined by pseudostratified respiratory epithelium **(Figure 1and2)** with benign subepithelial mucous glands **(Figure 3)**.

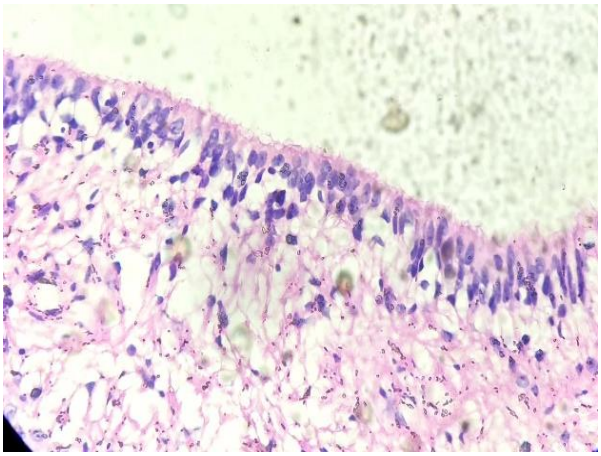

Figure 1: Hematoxylin and Eosin staining showed the cyst wall lined by pseudostratified respiratory epithelium

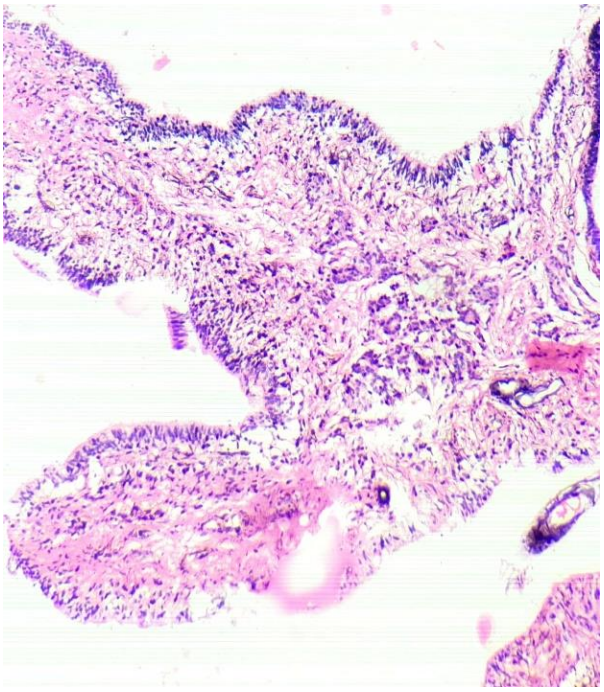

Figure 2: Hematoxylin and Eosin staining showed the cyst wall lined by pseudostratified respiratory epithelium

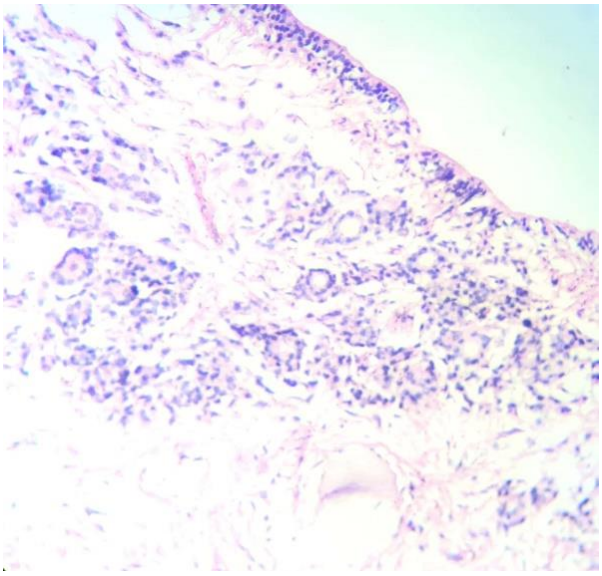

Figure 3: Benign subepithelial mucous glands

There was no evidence of glial tumor and granulomatous lesion. These features are of benign intraspinal intramedullary cyst having benign respiratory epithelium i.e. bronchogenic cyst.

DISCUSSION

Bronchogenic cysts are found most frequently along the tracheobronchial tree in the mediastinum or with the lung parenchyma. Rarely, these cysts have been found in other locations, including cutaneous[7] and subcutaneous tissues[8], the pericardium[9], the diaphragm[10], the abdomen[11], and the spinal cord.

Histologically, congenital intraspinal cysts can be classified into 3 types: epithelial, mesenchymal, and mixed types. Epithelial cysts can be further divided into endodermal (neurenteric and enterogenous) and ependymal cysts.[12] Endodermal cysts, which are uncommon lesions, account for 0.5% of spinal cord space-occupying lesions.[13] There were previous various views on the definition of endodermal cysts; however, it is defined the characteristics of typical endodermal cysts as follows: cysts have a gastrointestinal and/or respiratory epithelium, and mainly occurs in the cervical and thoracic regions.[14] The term bronchogenic cyst indicates tissues surrounded by the respiratory tract epithelium around endodermal cysts. So far the exact mechanism has not been found. Generally, it is considered that bronchogenic cysts are caused by anomalous embryological connections between the primitive foregut and developing neural tube and are related to splitting or reduplication of intervening notochords.[15]

Intraspinal cysts are categorised into three categories based on histological features.[16]

Category A–simple cyst lined by epithelium on a basement membrane with a thin wall of connective tissue

Category B–cyst lined by epithelium with a wall containing tissues found along the gastrointestinal tract or tracheobronchial tree.

Category C–cyst lined by epithelium with a wall containing ependymal and glial tissues as an intrinsic part of the lesion.

In our case, there was respiratory epithelium with cyst wall containing mucous glands so it can be considered under category B.

CONCLUSION

To conclude, bronchogenic cysts are very rare congenital malformations in the intraspinal area. Most of them are found in the intraduralextramedullary area. But ours is unusual case of intradural and intramedullary bronchogenic cyst (category B) in dorsal spine in an adult male patient .To best of our knowledge it is the third case report of its kind.

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