

CASE STUDY

Cervical Bronchogenic Cysts[☆]

Quiste broncogénico cervical

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Introduction

Bronchogenic cysts are benign congenital malformations derived from the tracheobronchial tree that correspond to non-functioning lung tissue.^{1,2} The majority of cases are diagnosed in the pediatric population.³

In adults they are fundamentally found in mediastinum and lung parenchyma.³ However, they can sometimes be located in the thoracic wall, abdomen or cervical region⁴; in the cervical area, they can be mistaken for another type of less common cervical lesion.

We present the case of an adult patient who presented with a cervical mass whose definitive diagnosis was bronchogenic cyst.

Clinical Case

This was a 67-year-old patient, without relevant history, who presented with fluctuating left laterocervical discomfort

that had lasted 10 years, without any other accompanying symptoms.

In the examination, a soft nodule could be felt, not adhered to deep planes; it was approximately 2 cm in diameter and in the left IIA area. The rest of the otorhinolaryngological (ORL) examination was normal.

The ultrasound revealed an adenopathic conglomerate mass at the level of the hyoid cartilage (some with calcifications inside).

A mass could be seen in the cervical CT: 26 mm × 20 mm, lobulated and hypodense, with some calcifications inside in the left submandibular area; filling absent in the left jugular vein in almost its entire path, suggesting vein thrombosis.

The cervical MR (Fig. 1) revealed a polylobulated, multiseptated cystic image, with faint capture of its thin 23-mm × 26-mm wall located opposite the jugular-carotid space, medial to the left sternocleidomastoid muscle. Likewise, thrombosis of the internal jugular vein was confirmed.

Because of the presence of a cervical lesion of suspicious origin, surgical excision and later pathological study were performed.

During the operation various rounded lesions with a gelatinous appearance were removed at the left cervical II and III levels.

The pathology study revealed the existence of several cystic lesions with walls formed of mucus-secreting cylindrical epithelium and of smooth muscle. It also found mucous

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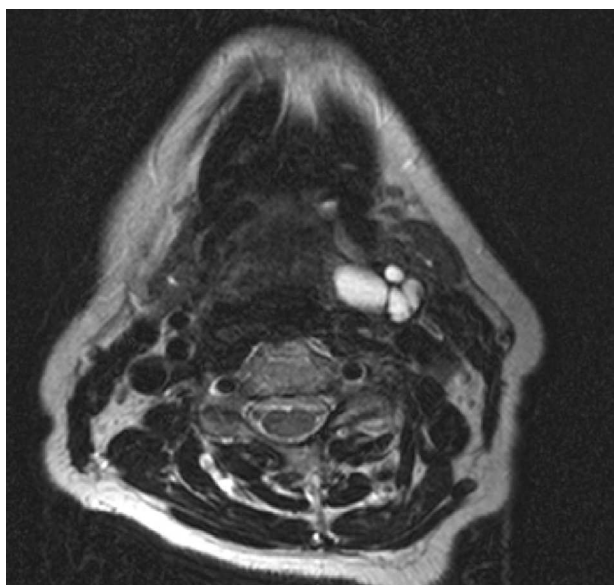


Figure 1 Cervical MR.

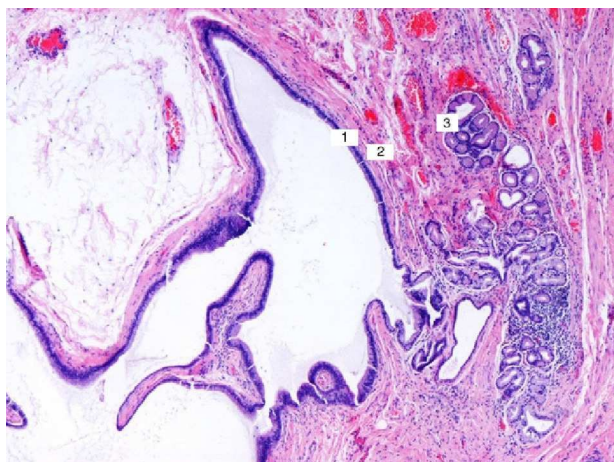


Figure 2 (H-E). Cystic lesion with wall formed of mucus-secreting cylindrical epithelium (1) and of smooth muscle (2), as well as mucous glands (3) within the thickness of the cyst wall.

glands within the thickness of the cyst wall, compatible with the diagnosis of bronchogenic cyst (Fig. 2).

The patient was discharged 72 hours after the operation without anticoagulant treatment, after being diagnosed with chronic jugular thrombosis by the Vascular Surgery Service.

At present the patient shows no recurrence of the lesions and cervical palpation is not pathological.

Discussion

Bronchogenic cysts are congenital malformations of the tracheobronchial tree that are normally found in the thoracic cavity, in relation to the bronchial tree, in the peripheral area of lung parenchyma or in the mediastinum.^{1,2,8} However, there are some cases in which these cysts are located

in the thoracic wall, mainly in the presternal subcutaneous cell tissue, in the cervical area or the abdomen.

It is postulated that bronchogenic cysts originate between the 5th and 7th week of gestation.⁴ These cysts are thought to form through alterations in the development of the primitive intestine, which splits in two: a ventral division then gives rise to the trachea, while the other, dorsal section is the origin of the esophagus.⁸ If the abnormal bud is resorbed, there are no problems; however, if it persists, it can lead to the development of a bronchogenic cyst.⁹ These buds are capable of migrating during the course of their development, with the possibility of settling into several intra- and extrathoracic locations.^{9,12}

Maier, in 1948, classified the bronchogenic cysts according to their location as paratracheal, carinal, hilar, paraesophageal and atypical (located in the diaphragm, abdomen, skin or subcutaneous tissues such as in the cervical area).^{2,5,9}

From approximately 70 published cases of cervical bronchogenic cysts, 75% were located in the midline of the neck, most commonly in the upper third, while the rest appeared in lateral lines, mostly in the lower third.^{2,9} This difference in placement may be due to the embryological development of the tracheobronchial tree. According to the literature, the cyst would be located in the midline if the bud occurred during the development of the trachea; while the location would be laterocervical if it originates during the development of the bronchial tree.⁹

Cervical bronchogenic cysts in adults generally present as an asymptomatic mass lacking apparent connection with the respiratory tree or the digestive tract.¹² However, if they are very large, they can produce dyspnea or dysphagia.³

They are typically soft, rounded or oval and have mucoid content. When the cysts get infected, it is usually the result of the existence of communication with the tracheobronchial tree.¹⁰

Because of this situation, various clinical diagnoses of more normal cervical lesions are postulated when there is a cervical mass of this nature, such as thyroid cyst, thyroglossal cyst, branchial cyst, dermoid cyst, lymphangioma, cystic hygroma, teratoma and cervical thymic cyst.¹⁻³

With respect to imaging tests, ultrasound gives us information as to the cystic nature of the lesion and its size. The CT helps us to better establish the location of the lesion and its relationship with other cervical structures; however, there are no specific radiological criteria that make it possible to suspect the presence of a bronchial cyst.⁸

The radiological test of choice would be MR, given its high resolution in the assessment of soft tissues, providing information on the relationship of the lesion with vital cervical structures. In the majority of cases, MR reveals a lesion with high intensity signal in T1 and T2, which is thought to be from the presence of high content of protein material; however, no specific radiological criteria exist for this type of pathology with this imaging test either.³

Likewise, no diagnosis can be established during the operation either, given that the mass simulates the more frequent congenital cysts.² In our case we suspected that the lesion could correspond to a cystic hygroma during the operation, due to its "gelatinous" appearance.

Definitive diagnosis requires pathological confirmation, which reveals a thin-walled cyst with an inner lining of pseudo-stratified cylindrical ciliated epithelium.⁴ It

contains mucous liquid or air, in addition to 1 or more bronchial elements (mucous glands, smooth muscle fibers and cartilage^{1,2}).

In the pathology laboratory, differential diagnosis should be performed with thyroglossal tract cysts and branchial cysts, which can have areas of respiratory epithelium but lack cartilage, smooth muscle tissue and seromucinous glands.⁴

As far as the finding of jugular thrombosis in our case, upon reviewing the literature we found the following as possible causes: lesion of the vascular wall (iatrogenic, from intravenous drug use or from cervical trauma), infections in the ORL zones, congenital or acquired hypercoagulable states (oral contraceptive use, antiphospholipid syndrome and paraneoplastic syndrome), venous stasis from compression secondary to tumors of the head and neck, mediastinum and idiopathic.^{5,6}

The classic recommendation as to the time anticoagulation should be maintained in deep venous thrombosis (DVT) is 3 months. In the last few years, various prospective studies have been carried out to evaluate the appropriate duration for anticoagulant treatment depending on the etiology of the thrombosis. Focusing on patients with a first DVT episode, it has been shown that the recurrence rate is significant less when the anticoagulant is administered for long period of time, with recurrence being similar in the group without anticoagulation and the group with anticoagulation for 6–12 months. Consequently, a treatment length of 3 months is recommended for patients with a first episode of venous thrombosis with transitory factors of risk.⁷

There is a general consensus in that bronchogenic cysts in adults should be treated with surgical removal, even if they are asymptomatic. In addition to confirming that the mass is benign, an early intervention can prevent future complications such as infections, compressive symptoms, the mass becoming malignant and the rare but serious gas embolism.^{11,12}

The potential that these lesions have of becoming malignant has been described in various cases of carcinoma that originated from bronchogenic cysts. The first case in the literature was reported by Moersch and Clagget in 1947, and since then there have been descriptions of various, well-documented cases of this type of pathology becoming malignant and turning into bronchoalveolar carcinoma, adenocarcinoma and even squamous cell cancer. No evidence has been provided to date on the carcinogenesis of these lesions, but it is postulated that instability in the epithelial cells of the cyst walls are responsible for them becoming malignant.¹³

Conclusion

Although cervical bronchogenic cysts are infrequent, we feel that they should be considered as part of the differential diagnosis for cervical lesions, both in those of laterocervical location and middle cervical location.

Conflict of Interests

The authors have no conflicts of interests to declare.

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