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경부절게를 통한 기관지원성낭종 치료

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Bronchogenic cysts treated with the transcervical approach

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= Abstract =

Bronchogenic cysts are congenital malformations of the bronchial tree, a type of bronchopulmonary foregut malformation. The presentation of the bronchogenic cyst is variable, making pre-operative diagnosis difficult. They aremostly asymptomatic orarefound incidentally when the chest is imaged. They can present as lower neck massesor mediastinal masses that may enlarge. They cause mass effect due to local compression and may result in tracheo-bronchial obstruction leading to air trapping and respiratory distress. The treatment is somewhat controversial, and in general,these lesions are treated using the transcervical or transbronchial approach. When these cysts arelocalized in the upper mediastinum, it may be possible to removethemusing the transcervical approach. In our three cases, the patientscomplained of mild dysphagia, foreign body sensation, and dyspnea. We report three cases of a large bronchogenic cyst in the lower neck and the upper mediastinum treated using the transcervical approach.

Key Words : Bronchogenic cyst, Dysphagia, Neck mass

Introduction

Bronchogenic cysts are rare congenital lesions accounting for only 5-10% of all paediatric mediastinal masses. They originate as sequestration from the ventral foregut, the antecedent of the tracheo-bronchial tree.[1,2] The cysts may usually lie within the mediastinum or the lung parenchyma. The incidence of mediastinal cysts is equal between the sexes, whereas intrapulmonary cysts are reported to have a male predilection.[3,4] The cyst wall is composed of an inner layer of ciliated respiratory epithelium with cartilage, mucous glands, smooth muscles, and fibrous tissue. Almost all cases are asymptomatic. Symptoms includechest pain, dyspnea, cough, stridor, and respiratory compromise when they cause tracheal/ bronchial compression. Symptoms vary with age at presentation, and to a large extent, they depend upon the size and location of the cyst.[3,5,6]

Chest X-ray findings suggest the possibility of a mass lesion in the mediastinum. A barium swallow study shows extrinsic compression of the esophagus and contrast-enhanced chest CT scan clinches the diagnosis and helps in taking the decision of surgical treatment. Endoluminal sonography and MRI are also helpful and highly sensitive and specific for the diagnosis of this condition. The treatment is somewhat controversial. Sometimes, transcervical aspiration under ultrasound or CT guidance is performed to confirm the

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diagnosis and to treat these lesions. Some authors suggest surgical excision of all cysts because of their tendency to become infected or to transform into malignancy.4,7 Generally, these lesions are treated using the transcervical or transbronchial approach. Small lesions can be followed; however, they do have a tendency to increase in size over time. Complications includefistula formation with the bronchial tree, ulceration of the cyst wall, secondary bronchial atresia, superimposed infection, haemorrhage, malignant transformation is very rare (0.7% risk), but reported, with primaries includingrhabdomyosarcoma.[5,8)]

These cases demonstrate the need for detailed investigations prior to treatment of patients with such a symptom complex as a bronchogenic cyst may be the cause of such symptoms.

Subjects

The first case, a 69-year-old woman, visited our hospital

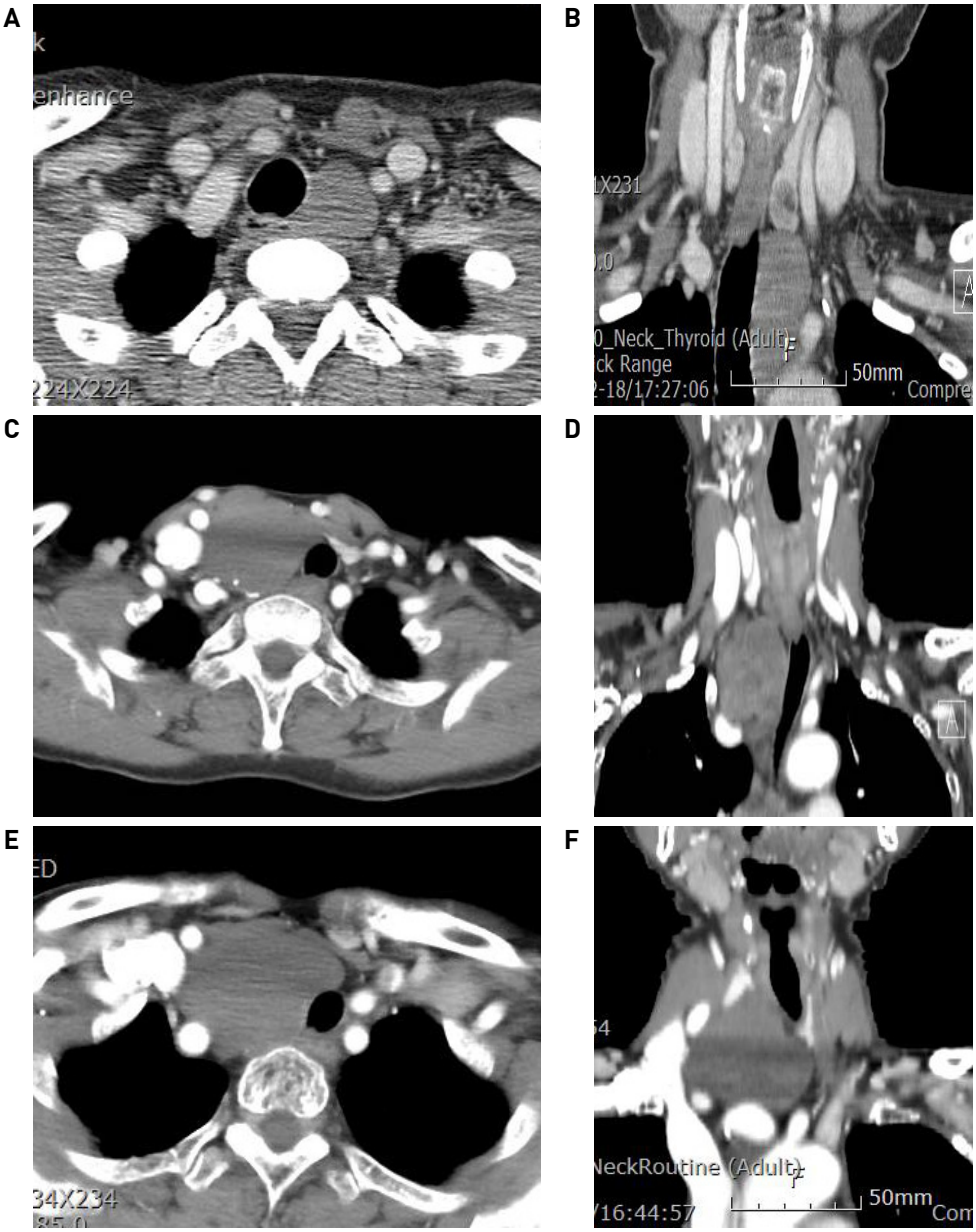

Fig. 1. Preoperative axial(A,C,E) and coronal(B,D,F) CT images for three cases. Left lower neck mass shows well defined cystic mass on lower portion of the left thyroid gland. It compresses the trachea mildly to the right side and involves upper mediastinum(A,B), Right cystic mass compresses the trachea severely to the left side and involves upper mediastinum(C,D), and more huge and more severely compresses the trachea severely to the left side and involves upper mediastinum(E,F). No enhancement of the mass suggests that the lesion is cystic.
CT; computerized tomogram

with a neck mass and foreign body sensation for 2 years. She complained of mild dysphagia. The neck mass, measuring 4x4 cm in size, was soft and freely/easily mobile, and it was located on the lower left neck. The patient did not complain of voice change and the vocal folds were freely/easily mobile on the laryngoscopic examination. She had undergone thyroid and parathyroid function tests and they showed normal values. Needle aspiration of the cystic mass was attempted, but the content was only water-like fluid. Ultrasonography of the neck revealed a huge cystic mass filled with fluid. The mass extended to the retrosternal area, and the trachea and the esophagus were displaced to the right side.(Fig. 1) Preoperative CT scan showed an approximately 3x6 cm in size, well- defined cystic mass on the lower portion of the left lobe of the thyroid gland which involved the upper mediastinum and caused tracheal deviation to the right side. Absence of enhancement of the mass suggested that the lesion was cystic. Also, there was calcification within the mass. Subsequently, she underwent neck exploration for thyroid and parathyroid surgery.(Fig. 1) A mass was found on the left lower pole of the thyroid gland and the upper mediastinum. The mass was dissected free from the surrounding tissues without adhesion. The thyroid gland and the recurrent laryngeal nerve on the right side were identified and preserved.Sectioning of the mass at the time of frozen section showed a thin-walled cyst containing a large amount of colorless watery fluid. Representative frozen and permanent histologic sections of the cyst wall demonstrated a thin and fibrous wall. Overall, the histopathological findings were consistent with a bronchogenic cyst. (Fig. 2)

The second case, a 51-year-old woman, presented with a swelling onthe right sternal area and foreign body sensation for 5 years. The neck mass was very soft and freely/easily mobile, and it was located on the lower right neck. The patient complained of mild dysphagia, but she did not complain of voice change. Ultrasonography and CT images showed a well-defined mass measuring approximately 3x4cm

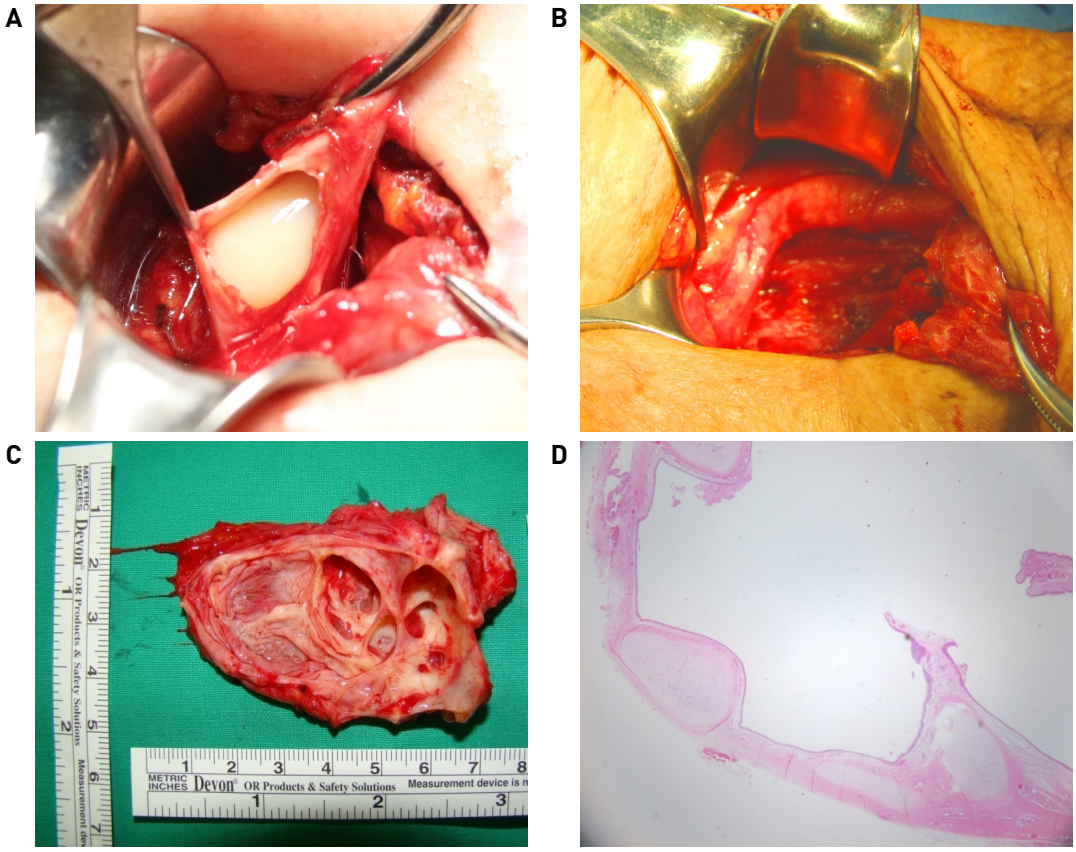

Fig. 2. Intraoperative findings. The cyst is filled with milky fluid(A), operative field after the complete excision of mass(B), a surgical specimen(C), smooth and multi-septated measuring about 7x4cm, and photomicrograph (H and E stain, original magnification ×25) of the cyst wall showing absence of epithelial lining with presence of discrete smooth muscle bundles in the cyst wall suggesting a bronchogenic origin for the cyst(D).
H and E: Hematocylin and Eosin

in size in the rightupper paratracheal area.(Fig. 1) It was a non-enhancing cystic mass, suggesting a thymic cyst or an esophageal duplication cyst. This mass extended almost equally on either side of the midline and was found to compress the upper esophagus posteriorly. An attempt was made to dissect the cyst from the surrounding structures. The mass was confirmed to have a cystic nature and it was filled with milky yellow fluid. There was absence ofdense adhesions and immense vascularity and the mass was easily dissected without any difficulty.

The third case, a 74-year-old woman, presented with a swelling on the right lower neck and the sternal area, dysphagia, and mild dyspnea for 6 months. The neck mass was very soft and freely/easily mobile, and it was located on the lower right neck.The CT images showed a well-defined mass measuring approximately 5x4cm in size in the right lower neck and the upper mediastinum. This mass severely compressed the esophagus and the trachea. During surgery, the cyst was dissected from the surrounding structures.(Fig. 1) The mass was confirmed to have a cystic nature and it was filled with water.

In all the above three cases, the cysts were excised successfully via the transcervical approach and the representative tissues were sent for histopathological examination. The bed of the excised area was ablated with electrocautery. The patients had an uneventful recovery and were discharged on the 3rd post-operative day. Histological evaluation confirmed the diagnosis of a bronchogenic cyst.

Discussion

Bronchogenic cysts are rare congenital malformations of the bronchopulmonary foregut and they account for only 5-10% of all paediatric mediastinal masses. They form as a result of abnormal budding of the bronchial tree during embryogenesis (between the 4th-6th week). 3,5Bronchogenic cysts are lined by secretory respiratory epithelium;cuboid or columnar ciliated epithelium. The wall is made up of tissues similar to those of the normal bronchial tree, including cartilage, elastic tissues, mucous glands, and smooth muscle.The incidence of mediastinal cysts is equal between the sexes, whereas intrapulmonary cysts are reported to have a male predilection.They can occur in the mediastinum or be intrapulmonary, andthe distribution of locations can be quite varied and they are rarely multiple.[6,7] The most common location is the middle mediastinum (65-90%). Mainly, bronchogenic cysts are asymptomatic and are found incidentally when the chest is imaged. They can present as mediastinal masses that may enlarge and cause local compression. When they are large, mass effect may result in bronchial obstruction leading to air trapping and respiratory distress. An alternative presentation may occur when the cyst becomes infected.[8]

Bronchogenic cystsmainly contain water, and sometimes, they may containvariable amounts of proteinaceous material, blood products, and calcium oxalate. The latter three components result in increased attenuation mimicking solid lesions. They do not usually communicate with the bronchial tree, and are typically not air filled. Although bronchogenic cysts are usually fluid filled, occasionally a communication may develop following infection, resulting in an air-filled cystic structure or an air-fluid level. 4-6 On CT images, the cysts usually appear as soft-tissue density rounded structures, sometimes with compression of the surrounding structures. Such compression can lead to air trapping and a hyperlucent hemithorax. They appear as well-circumscribed spherical or ovoid masses of variable attenuation with variable fluid composition explaining the different CT attenuations observed.Thus,a significant proportion of masses are of soft tissue densityor even hyperdense to surrounding mediastinal soft tissues. CT is better able to detect calcium oxalate (milk of calcium) layering dependently. Usually, there is no solid contrast enhancement. MRI is occasionally performed for confirmation.T1-weighted images show variable signal intensity, from low (similar to fluid) to high (due to protein content)fluid levels, attributed to layering of variable fluid content. T2-weighted images usually show high signal intensity due to fluid content.[5,6]

Appropriate treatment of patients with bronchogenic cysts depends on the patient's age and symptoms at presentation. Conservative treatment has been advocated in asymptomatic or other high-risk patients. In selected cases, percutaneous catheter drainage or sterile alcohol ablation can be performed. However, it is generally recommended that asymptomatic cysts in young patients be removed because of the low surgical risk and the potential risk of late complications (albeit rare) such as infection, hemorrhage, or neoplasia within the cyst. Symptomatic cysts are generally resected by means

of thoracotomy or video-assisted thoracoscopy, but they can be resected via the transcervical approach when localized in the upper mediastinum. Special attention should be paid to avoid injury to the vagus nerve, the tracheo-bronchial tree, pericardium, and the esophagus. When the cyst is located in the upper mediastinum, it is possible to remove the cyst successfully via the transcervical approach. However, when the cyst is located within the mediastinum, it should be approachedby thoracoscopic/videoscopic resection.[3-7)]

In conclusion, bronchogenic cysts are rare tumors in the mediastinum. In our three cases, patientscomplained of mild dysphagia, foreign body sensation and dyspnea. When they are located in the upper mediastinum, we recommend mass removal by making atranscervical incision. It is important to identify the location of the mass with use of contrast-enhanced chest CT scan. Otherwise, the surgical option should be considered a definitive procedure in all cases to avoid the development of complications.

중심 단어 : 기관지원성낭종. 경부절제

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