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The Occurrence of a Branchial Cleft Cyst in the Anterior Mediastinum: A Case Report[1]

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Branchial cleft cysts and branchial anomalies develop from the branchial cleft apparatus that persists after fetal development. The most common anatomical site for the occurrence of branchial cleft cysts is in the cervical area, generally anterior to the sternomastoid muscle in the upper or middle portion of the neck. A mediastinal branchial cleft cyst is extremely rare and few cases have been reported. We report the case of branchial cleft cyst found in the anterior mediastinum with literature review.

Index words : Branchioma
Mediastinal cyst
Tomography, X-Ray Computed
Thorax

Branchial cleft cysts are congenital anomalies which are usually expressed in the form of a cystic mass in the neck. Anomalies of the second branchial cleft account for approximately 95% of all branchial cleft cyst (1). Congenital abnormalities of the branchial apparatus can result in various abnormal conditions of the neck including cyst, sinus, or fistulae. The number of cysts is more common than sinuses and fistulae combined. Moreover the anomalies are rarely bilateral and can be familial (1). Despite being congenital, cysts are generally found in young adults (average age, 31 years) as a mass at the mandibular angle, often associated with an upper respiratory tract infection resulting from an injury, which subsequently precipitates towards cyst enlargement (1). To the best of our knowledge, only two cases of mediastinal branchial cleft cysts have been reported in the literature (2, 3).

Case Report

A 56 year-old man was admitted to the hospital for a health screening. A chest film revealed well defined mediastinal mass adjacent to right cardiac border (Fig. 1A). A chest CT revealed a 5.5×5.0 cm non-enhancing cystic lesion located at the anterior mediastinum. In addition, the heart was found to be deviated to the right. The cyst revealed a well defined lobulated thick wall with no evidence of an abnormal enhancing lesion within the cyst (Fig. 1B). Consequently, a diagnostic thoracotomy was performed to obtain an excision biopsy. The mass was found to be smooth and firm with the gross specimen showing cystic mass surrounded by adipose tissue measuring 8.5×5.0×4.0 cm (Fig. 1C). The unicystic lesion had inner smooth surface and chocolate colored cystic fluid. Sections of cystic mass from mediastinum showed a thick collagenous wall with a single layer of ciliated columnar branchial epithelium. In addition, prominent lymphoid aggregations was observed on the fibrous wall. The outer surface of the cystic lesion was sur-

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rounded by mature adipose tissue with no evidence of thymic tissue or Hassall's corpuscles within. As a result, this finding excludes the possibility of a thymic cyst. All histological sections were reviewed by two pathologists. The final pathologic diagnosis was determined to be a branchial cleft cyst.

Discussion

Mediastinal cysts are uncommon lesions that comprise approximately 10 - 27% of all mediastinal masses. Moreover, they are believed to primarily be a result of developmental anomalies of the foregut or related embryonal structures (4). Congenital mediastinal cysts are classified as bronchogenic, duplication, neurenteric, pericardial, and thymic (5). The CT features of benign mediastinal cysts are smooth, oval or tubular with a well-defined thin wall that usually enhances with a homogeneous attenuation range of water attenuation, no enhancement of the cyst contents, and no infiltration of the adjacent mediastinal structure (5). The diagnosis of the mediastinal cysts is attained by examining a patient's clinical history and manifestations, as well as the anatomic position or certain features seen with the CT

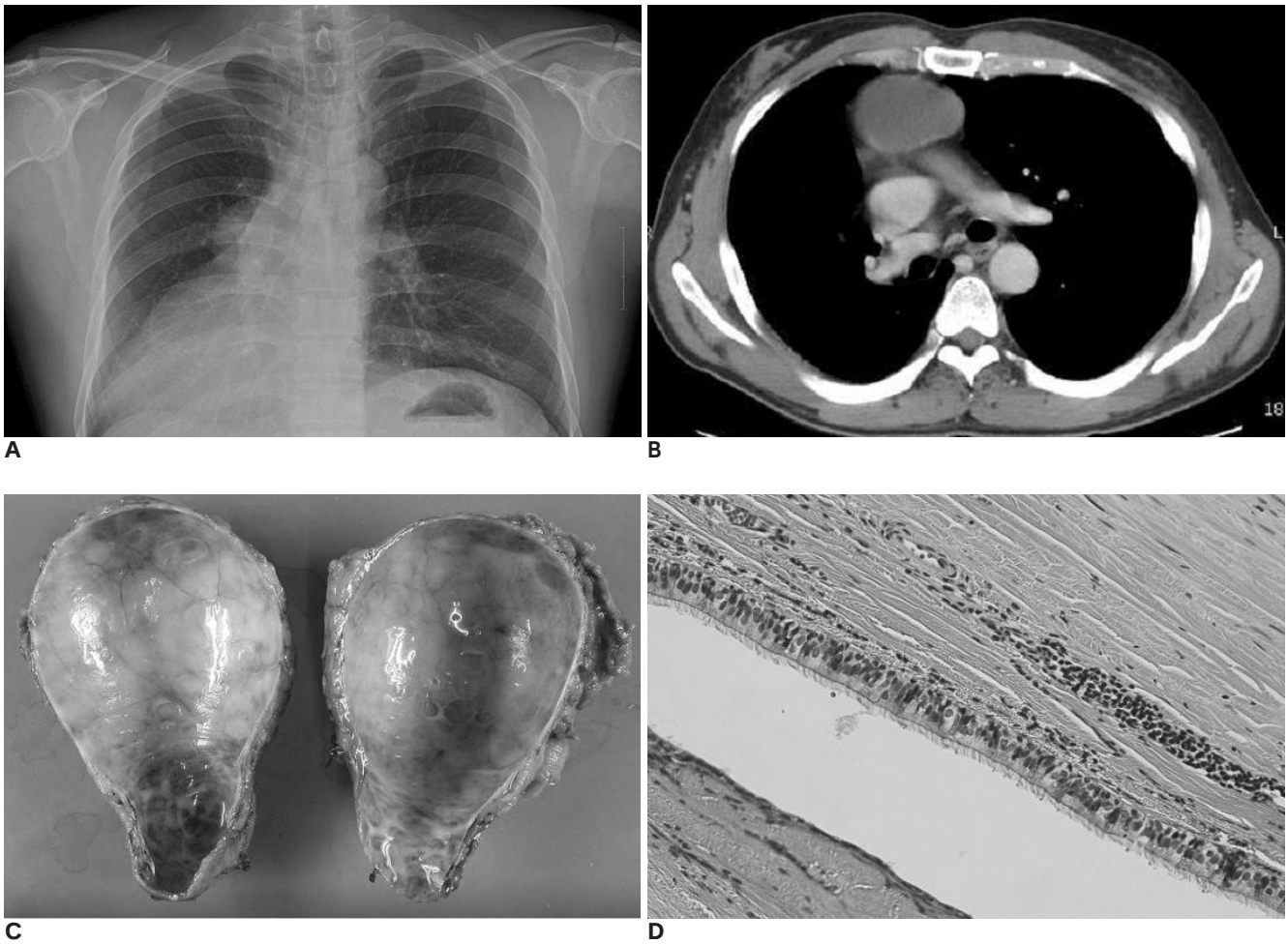

Fig. 1. A 56-years-old man with anterior mediastinal branchial cleft cyst.
A. A chest radiograph revealed a well defined mediastinal mass (arrow) causing a right deviation of the heart.
B. A well defined anterior mediastinal cystic mass with a thick wall was observed at the anterior portion of the pulmonary trunk. No evidence of abnormal enhancing lesions was found within the cyst. The right side heart seemed to be dextrocardia; however, the aorta originated from the left ventricle and the pulmonary artery originated from the right ventricle without any other anomalies of the heart.
C. The gross specimen revealed a cystic mass surrounded by adipose tissue, which measured 8.5×5.0×4.0 cm. The unicystic lesion had an inner smooth surface and a chocolate colored cystic fluid with a thick wall.
D. A thick collagenous wall and a single layer of ciliated columnar branchial epithelium was identified along with a prominent lymphoid aggregating at the fibrous wall. No evidence of thymic tissue or Hassall's corpuscles was observed within the cysts (H & E, ×200).

which are important for the correct diagnosis of malignancy.

Embryologic anomalies of the branchial arches are manifested as cysts, fistulae, and sinuses. Knowledge of the embryologic basis of these defects helps one understand these anomalies. By the end of the fourth week of embryonic life, the branchial arches and the mesenchyma are easily identified (6). Six pairs of branchial clefts are separated by six aortic arch arteries. The first two pairs of arch arteries are involuted, whereas the third pair form the common and proximal portion of the internal carotid artery. The fourth pair forms the right subclavian artery with the left arch becoming part of the aortic arch. Lastly, the fifth pair of arch arteries involute, whereas the sixth pair forms the pulmonary arteries (7). The first branchial cleft normally gives rise to the Eustachian tube, tympanic cavity, and mastoid antrum. This contributes to the formation of the tympanic membrane. It is the only cleft to contribute to an adult structure (external auditory canal). The second, third, and fourth branchial clefts are part of an ectodermal lined depression known as the cervical sinus of His. As the second and fifth branchial clefts merge with each other, the cervical sinus of His is obliterated. The second branchial pouch, lined by endoderm, gives rise to the palatine tonsil and tonsillar fossa. The third branchial pouch forms the inferior parathyroid gland, thymus, and pyriform sinus; whereas, the fourth branchial pouch leads to the formation of the superior parathyroid gland and apex of the pyriform sinus (6).

Generally branchial cleft cysts are located on the lateral neck; however,the mediastinum would need to be involved in order for a cyst to arise from the fourth or fifth branchial cleft (2). Because the fourth, fifth, and sixth branchial clefts are located below the fourth aortic arch artery during the embryologic stage, the cyst would course below the fourth branchial artery or its derivation, namely, the aortic arch or the right subclavian artery (2). Theoretically, the existence of fifth or sixth branchial clefts and branchial cleft anomalies are possible in humans. They would course below the aortic arch or right subclavian artery and then the branchial cleft cyst located at the mediastinum (below fourth aortic arch artery). William et al. suggests that mediastinal branchial cleft cysts originated from fourth or fifth branchial cleft (2).

The classical branchial cleft cyst is characterized histologically by an epithelial lining which is typically squamous, but may be ciliated columnar. Underlying the epithelium is abundant lymphoid tissue with germinal centers (2).

The radiologic feature of the mediastinal branchial cleft cysts exhibit no specific findings to distinguish them from other anterior mediastinal cysts, which include thymic or bronchogenic cysts. Pathologically it is important to distinguish a branchial cleft cyst from a thymic cyst because the thymic cyst would be lined by ciliated columnar epithelium. The correct diagnosis of thymic cysts can be established by the detection of thymic tissue and Hassall's corpuscles within the cyst (8). In our case study, no thymic tissue or Hassall's corpuscles were identified. Moreover, no characteristic radiologic findings were seen to permit us to differentiate mediastinal branchial cleft cyst from other types of anterior mediastinal cysts (bronchogenic cysts or thymic cysts). However, a branchial cleft cyst should be considered and the differential diagnosis of anterior mediastinal cysts should be outlined.

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전종격동에서 발생한 아가미틈새물혹: 증례 보고[1]

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아가미틈새물혹과 아가미틈새기형은 태생 기에서 아가미틈새기관의 흔적기관이 퇴화되지 않고 남아서 발생하는 선천성 질환이다. 아가미틈새물혹은 목의 중상부에 있는 흉쇄유돌근의 앞쪽에서 흔히 나타난다. 종격동의 아가미틈새물혹은 극히 드물며 극소수의 증례만이 보고되어 있다. 이에 저자들은 전종격동에서 발생한 아가미틈새물혹을 문헌 고찰과 함께 보고하는 바이다.