
A case of cervical bronchogenic cyst presenting with hoarseness in an adult

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ÖZET

Erişkinde ses kısıklığı ile kendini gösteren bir servikal bronkojenik kist olgusu

Servikal kitlesi olan 28 yaşında erkek hasta, ses kısıklığı ve kilo kaybı şikayetleriyle kliniğimize sevk edildi. Bilgisayarlı göğüs tomografisinde suprajuguler çentikte yerleşmiş 3 x 4 cm'lik kistik bir lezyon görüldü. Servikal ultrasonografi lezyonun kistik olduğunu teyit etti. Kulak burun boğaz muayenesinde sağ vokal kord paralitikti. Perkütan iğne biyopsisi tanı sağlamadı. Suprajuguler insizyon ile kist tamamen çıkarıldı. Histopatolojik değerlendirme lezyonun bir bronkojenik kist olduğunu gösterdi. Hastanın ses kısıklığı kistin çıkarılmasından hemen sonra düzeldi. Hasta postoperatif üçüncü günde taburcu edildi. Altıncı ayda yapılan kontrolünde nüks tespit edilmedi.

Anahtar Kelimeler: Bronkojenik kist, boyun, ses kısıklığı.

SUMMARY

A case of cervical bronchogenic cyst presenting with hoarseness in an adult

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A twenty-eight year old male with a cervical mass, complaining of hoarseness and weight loss, was referred to our clinic. A 3 x 4 cm cystic lesion localizing in the region of the suprajugular notch was seen with the aid of computerized chest tomography. A cervical ultrasound confirmed that it was a cystic lesion. It was discovered that the right vocal cord was paralytic after an ear-nose-throat examination. A percutaneous needle biopsy did not provide a diagnosis. The cyst was completely excised via suprajugular incision. Histopathologic examination showed that it was a bronchogenic cyst. The hoarseness was completely eliminated immediately after cystic removal. The patient was discharged after the third postoperative day. After a six-month of follow-up, no recurrence was detected.

Key Words: Bronchogenic cyst, neck, hoarseness.

Bronchogenic cyst (BC) is a rare congenital lesion with a reported prevalence of 1/42.000 to 1/68.000 of population (1). In a recent study a series of 2.163 mediastinal lesions, 72 (3.3%) were found to be due to BCs (1). During embryogenesis, the primitive foregut arises. The laryngotracheal groove develops from the ventral wall of the primitive foregut and then forms the trachea and the bilateral bronchial buds. Further budding results in the development of the bronchial trees. It has been hypothesized that BCs originate from abnormal budding (2).

The majority of BCs are located in the mediastinum and the lung parenchyma; however, ectopic locations have also been described: These include paratracheal, cutaneous, lingual, supraclavicular, diaphragmatic, intrapericardial, spinal, abdominal areas, and the neck (3). BCs may be intrathoracic or extrathoracic. Extrathoracic cysts commonly occur in the suprasternal notch or manubrium sterni: Other sites being the neck, chin, base of the tongue, shoulder and the scapular region. The male/female ratio is 4/1 (4).

We herein reported an interesting case of cervical BC with a very rare symptom of hoarseness due to recurrent nerve palsy resulted from cystic compression in an adult patient. In addition, cyst characteristics and histological findings of this lesion were also discussed.

CASE REPORT

A 28-year-old man was referred to our clinic for an evaluation of a cervical mass in the suprasternal notch, which had been first noticed two weeks prior to our examination. Six days before our evaluation, the patient had been experien-

cing hoarseness. His medical history included a healthy childhood, no history of recurrent upper respiratory infections, dyspnea or dysphagia. Upon physical examination, a soft elastic mass, approximately 3 x 4 cm in size, was identified above the jugular notch. Hematological evaluations, blood chemistry, and thyroid function tests were normal. A chest X-ray revealed no abnormal findings in the mediastinum and intrapulmonary regions. Computerized tomography (CT) of the chest revealed a single cyst 3-4 cm in diameter in the suprasternal region, beginning at level C7, and ending at level T1 (Figure 1). Cervical ultrasound revealed a 37 x 22 x 45 mm subcutaneous hypoechoic cystic mass in the antero-medial region of the neck. Diagnostic percutaneous needle aspiration was conducted in the region of the cyst and 2 mL of white mucoid material was aspirated. After aspiration of the cyst, hoarseness was transiently improved; however, it recurred one week later. The cytological examination concluded the mass was composed of a non-specific amorphous eosinophilic

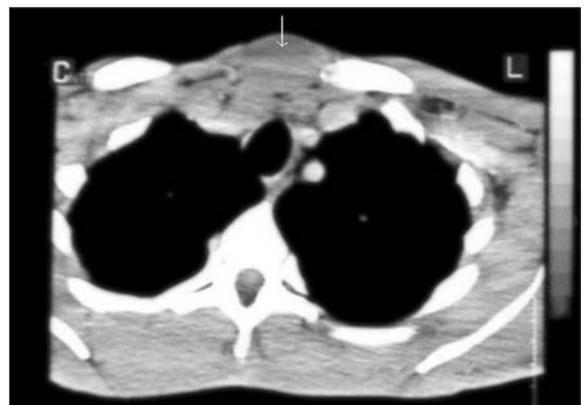


Figure 1. CT image showing a cystic lesion in the suprasternal region (white arrow).

cystic material. Tuberculosis culture and Ziehl-Nielsen stain were negative. Non-specific culture was not performed. The right vocal cord was found to be paralytic during the ear-nose-throat examination. A surgical excision of the mass, located in the subcutaneous area 1 cm below to 3 cm above the jugular notch, was carried out with a collar incision.

Microscopically, the cyst wall was layered with pseudostratified ciliated columnar epithelium. Dense mononuclear cell infiltration and mucous gland formation in the walls of the cyst were also seen (Figure 2). The histological diagnosis was reported as a BC.

After the surgical excision, hoarseness immediately disappeared. The patient was discharged on the third postoperative day. After a six-month follow-up, no recurrence was detected.

DISCUSSION

Localization of BCs in the cervical area is rare and suggestive of a developmental anomaly that had taken place before the fifth week of development. Cervical BCs are usually diagnosed in the paediatric population as an asymptomatic cervical mass. Although they are rarely observed in adults, they do reveal themselves through chest pain in the retrosternal area, dysphagia, cough, fever, hemoptysis, and recurrent infections (5). We found only one case that reported a BC causing hoarseness in the literature. Maung et al. re-

ported an instance in which multiple cervical BCs adjacent to the larynx caused hoarseness in the voice of the presenting individual (6). Our case can be distinguished from the later instance as he had a single lesion localized in the suprajugular notch. We believe that, the vocal cord paralysis was due to acute compression to the nerve. Evidence to support this conclusion was supplied by the fact that when the cystic fluid was aspirated hoarseness was transiently improved. However, one week later, his hoarseness recurred: Most probably due to refilling of the cyst with the fluid.

When we encounter a cystic tumour in the cervical region of the neck, we should consider its presence a result various diseases. These diseases are divided into two groups based on their original site. If the tumours have originated in the cervical region of the neck, we should consider papillary carcinomas of the thyroid with cystic changes, follicular adenomas of the thyroid, bronchial cysts, lymphangiomas, and dermoid cysts (7). On the other hand, mediastinal tumours with cystic lesions may be diagnosed as one of the following diseases: Tracheal diverticular formations, lymphoepithelial cyst, laryngocele, pharyngocele, Zenker diverticula, apical pulmonary hernia, thyroglossal duct cysts, cystic teratoma, branchial cleft cysts, epidermoid cysts, Hodgkin's disease, thyroid adenoma, thyroid cysts, and thymic cysts (1,2).

The definitive diagnosis of BCs can only be confirmed by a histopathological examination. They are characteristically lined with a pseudostratified ciliated columnar epithelium accompanied by underlying seromucinous glands. They can be observed on chest roentgenograms and CTs as smoothly bordered, oval, or round lesions with a homogeneous consistency or air-liquid level. They are generally not connected to the airways (8). Connections between BCs and the tracheobronchial system or esophagus are rarely observed and usually occur with intrapulmonary cysts. Air within the cyst is uncommon and suggestive of secondary infection and connection with the tracheobronchial tree. Calcification occurs occasionally in the wall or within the contents of the cyst (9).

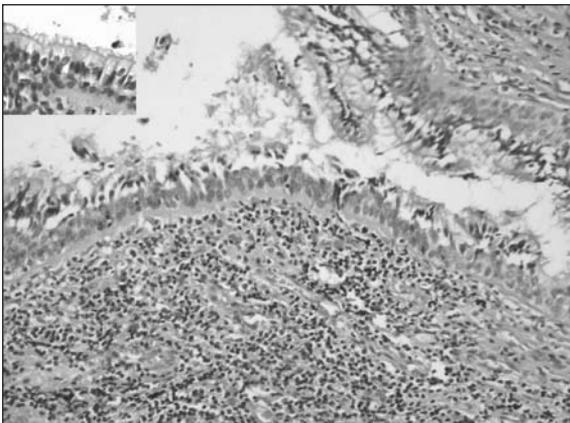


Figure 2. The cyst is lined by pseudostratified ciliated columnar epithelium. Lenfoid tissue is observed beneath the epithelium. Inset shows pseudostratified ciliated columnar epithelium (HE, x20).

In the presence of these conditions and with the risk of malignant transformation, complete excision is the only choice of treatment (3). No recurrence has been described when a complete surgical excision is carried out. Aspiration of the cyst is an inadequate treatment because recurrence after this procedure occurs (10).

In conclusion, cervical BCs are rare lesions and may cause hoarseness, which is an extremely unusual symptom, resulting from cystic compression. Based on our observations, when such lesions in the cutaneous or subcutaneous region are detected, a surgical excision should be conducted to reach an exact histopathological diagnosis for appropriate therapy.

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