Case Report / Olgu Sunumu



A rare cause of neck swelling: Spontaneous bleeding of the bronchogenic cyst

Boyundaki şişliğin nadir bir nedeni: Bronkojenik kistin spontan kanaması

Yeşim Esen Yiğit¹, Yaşar Kemal Duymaz¹, Ahmet Adnan Cırık¹, Büşra Balcıoğlu¹, Halil Çiftçi²

¹Department of Otolaryngology, Health Sciences University, Ümraniye Training and Research Hospital, Istanbul, Türkiye ²Department of Thoracic Surgery, Health Sciences University, Ümraniye Training and Research Hospital, Istanbul, Türkiye

ABSTRACT

Neck swellings can be caused by a variety of conditions, such as hemorrhage, congenital lesions, reactive lymphadenopathies, and cancer. Bronchogenic cyst is one of the congenital lesions of the lower airway. Bronchogenic cysts could expand and result in pain, compressive symptoms, and respiratory difficulty. A 45-year-old female presented to the emergency department with neck swelling, breathing abnormality, and pain. It was noted in the medical history that the patient regularly used aspirin. In the patient's examination and imaging studies, a 45×40×56 mm, well-contoured, nonenhancing cystic lesion was observed in the right paratracheal area extending to the neck. The lesion deviated the trachea to the left. During inpatient follow-up, the patient's complaints became markedly worse, and the patient had increased shortness of breath. Surgery was planned to remove the cyst, and the cyst was excised with the help of the thoracic surgery department. The pathology confirmed a bronchogenic cyst. In a patient experiencing neck swelling and respiratory distress, lesions spontaneously bleeding due to aspirin should be kept in mind. A fatal outcome may occur due to pressure on the airway.

Keywords: Antiaggregant use, anticoagulant use, aspirin use, bronchogenic cyst, neck swelling, respiratory distress, spontaneous bleeding, trachea deviation.

ÖΖ

Boyundaki şişlikler, kanama, konjenital lezyonlar, reaktif lenfadenopatiler ve kanser gibi çeşitli nedenlerden kaynaklanabilir. Bronkojenik kist, alt hava yolunun konjenital lezyonlarından biridir. Bronkojenik kistler genişleyebilir ve ağrı, bası semptomlarına ve solunum sıkıntısına yol acabilir. Kırk bes yasında kadın hasta boyunda şişlik, nefes almada zorluk ve ağrı ile acil servise başvurdu. Tıbbi öyküsünden hastanın düzenli aspirin kullandığı öğrenildi. Hastanın muayenesi ve görüntüleme incelemelerinde sağ paratrakeal alanda boyna uzanan 45×40×56 mm düzgün sınırlı, kontrast tutmayan kistik lezyon görüldü. Lezyonun trakeyi sola deviye ettiği görüldü. Hastanın yatarak takibinde yakınmalarının belirgin şekilde kötüleştiği ve nefes darlığının arttığı görüldü. Kistin çıkarılması için cerrahi planlandı ve göğüs cerrahisi bölümünün yardımıyla eksize edildi. Patoloji sonucu bronkojenik kisti doğruladı. Boyun şişliği ve solunum sıkıntısı ile karşılaşılan bir hastada aspirin nedeniyle spontan kanayan lezyonlar akılda tutulmalıdır. Havayoluna bası nedeniyle ölümcül bir sonuçla karşılaşılabilir.

Anahtar sözcükler: Antiagregan kullanımı, antikoagülan kullanımı, aspirin kullanımı, bronkojenik kist, boyun şişliği, solunum sıkıntısı, spontan kanama, trakea deviasyonu.

Bronchogenic cysts are rarely encountered; nevertheless, they are one of the most common malformations of the lower respiratory tract.^[1] They arise from anomalous budding of the foregut during development. Symptoms frequently appear in the

Received: November 14, 2023 Accepted: February 20, 2024 Published online: February 29, 2024

Correspondence: Yeşim Esen Yiğit, MD. **E-mail:** yyigit14@ku.edu.tr **Doi:** 10.5606/kbbu.2024.48379 third or fourth decade of life. Bronchogenic cysts can present with different symptoms. Esophageal bronchogenic cyst hemorrhage can result in esophageal obstruction.^[2] Pulmonary cysts can present as hemothorax.^[3] Mediastinal cysts can become infected.

Citation:

Esen Yiğit Y, Duymaz YK, Cırık AA, Balcıoğlu B, Çiftçi H. A rare cause of neck swelling: Spontaneous bleeding of the bronchogenic cyst. KBB Uygulamaları 2024;12(1):44-46. doi: 10.5606/kbbu.2024.48379.



Esen Yiğit Y et al. Spontaneous bleeding of the bronchogenic cyst

Furthermore, spontaneous bleeding inside a lesion may occur due to anticoagulant or antithrombotic use.^[4] The cyst may enlargen and cause obstructive symptoms, respiratory distress, and pain.^[5]

Neck swelling may occur due to multiple reasons, such as bleeding, congenital lesions, reactive lymphadenopathies, and malignancies.^[6] Herein, we report a case of mediastinal bronchogenic cyst exhibiting shortness of breath and neck swelling due to spontaneous bleeding inside the cyst.

CASE REPORT

A 45-year-old female presented to the emergency clinic with shortness of breath and neck swelling. The patient had neck pain for a few days. The pain had become unendurable in the last three days. The previous day, the patient had visited the interventional radiology clinic due to the neck swelling, and 50 mL of hematoma was drained. At the emergency clinic, the patient's vitals were stable. The patient's computed tomography (CT) and magnetic resonance imaging showed a 6x6 cm lobulated hypodense lesion at the right side of the neck reaching into the right intrathoracic region (Figures 1, 2). The lesion compressed the trachea to the left side and was adjacent to the esophagus. The thyroid appeared enlarged. The patient was later referred to our otolaryngology clinic. On inspection, the patient had marked neck swelling but no ecchymosis. On flexible endoscopic examination, the right vocal cord was paralyzed. The trachea appeared to deviate to the left side. The patient was hospitalized in the otolaryngology clinic to follow the airway condition.

On detailed history, a fine needle aspiration biopsy was performed due to a suspicious thyroid nodule (right lobe) 45 days ago. A previous ultrasound record showed that there was a 4 cm cystic lesion adjacent to the right lobe of the thyroid in the posteroinferior part. Previous thorax CT reports demonstrated a $45\times40\times56$ mm, well-contoured, nonenhancing cystic lesion in the right paratracheal area that displaced the trachea to the left. It was noted that the patient had diagnoses of arrhythmia, asthma, and thalassemia minor. The patient's regular medications were a beta-blocker and aspirin.

During follow-up, the complaints became markedly worse. A thoracic surgery department consultation



Figure 2. Magnetic resonance imaging of the bronchogenic cyst.



Figure 1. Computed tomography scan of the bronchogenic cyst.



Figure 3. Thorax computed tomography five months after surgery.

was carried out. The thoracic surgeon planned a thoracotomy, and the otolaryngology team would be involved if a problem with the neck were to occur. The patient was tried to be ventilated with a mask and intubated for the surgery. However, due to the right vocal cord paralysis, neck swelling, and potential tracheal deviation, the patient could not be ventilated and intubated. As the otolaryngology team, we performed an emergency tracheotomy. Later, the patient was intubated through the tracheostomy. Removal of the cyst was performed. Pathology revealed large-diameter fibrovascular structures involving neural structures, and the central part consisted of cystic structures. The bronchogenic cyst and tracheal deviation were absent in the imaging when the patient came for a control appointment after five months (Figure 3).

DISCUSSION

Complications of bronchogenic cysts include recurrent infection, compression of the gastrointestinal tract, compression of the heart and great vessels, rupture of the cyst, and malignant transformation.^[7] They can result in airway obstruction either due to their mass effect or cyst bleeding.^[5] In this case, the bronchogenic cyst bled spontaneously in a subacute setting due to aspirin use.

Differential diagnoses of neck swellings include reactive neck lymphadenopathy, congenital cysts, thyroid nodules, salivary gland tumors, tumors of the parapharyngeal space, malignant lymph nodes due to squamous cell carcinoma of the head and neck, metastases, or lymphoma. Bleeding may occur due to head and neck cancer and radiation therapy. As in our case, spontaneous bleeding inside an existing lesion may cause a hematoma in the neck. Spontaneous bleeding of the head and neck due to anticoagulant use has been reported.^[4,8] These were mostly laryngeal, retropharyngeal, and sublingual.^[4] Spontaneous bleeding of the parathyroid and thyroid gland has been reported, and it should be kept in mind when a patient presents with neck swelling. ^[9] A lesion of the thorax that caused hematoma and extended to the neck has been reported.^[10]

In conclusion, when a patient presents with neck swelling and has a history of an existing bronchogenic cyst and aspirin use, it must be kept in mind that the cyst may have hemorrhaged spontaneously. This may be life-threatening due to the airway compromise.

Patient Consent for Publication: A written informed consent was obtained from the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Fikir kavram: Y.E.Y., Y.K.D., H.Ç.; Tasarım: Y.E.Y., B.B.; Kontrol denetim: A.A.C., H.Ç.; Veri toplama/işleme: Y.E.Y.; Analiz/yorumlama: Y.E.Y., B.B.; Literatür taraması: B.B., A.A.C., Y.K.D.; Makalenin yazılması: Y.E.Y., Y.K.D.; Referanslar ve fonlar: A.A.C., H.Ç., Y.E.Y.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

REFERENCES

- Rodgers BM, Harman PK, Johnson AM. Bronchopulmonary foregut malformations. The spectrum of anomalies. Ann Surg 1986;203:517-24. doi: 10.1097/00000658-198605000-00011.
- Liang JS, Yin GL, Zhang XM, Zhu SB, Dong YQ. Intramural esophageal bronchogenic cyst with wall hemorrhage results in acute esophageal obstruction: Report of two cases. Am Surg 2014;80:E27-9.
- Suzuki T, Kamata S, Sato N. A case of massive hemothorax associated with an intrathoracic bronchogenic cyst. Gen Thorac Cardiovasc Surg 2020;68:396-8. doi: 10.1007/ s11748-019-01145-z.
- Thomas SA, Moideen SP, George SM, Ipe S, Menon DG. Management of spontaneous neck hematoma in patients on anticoagulant therapy: Our experience from a tertiary care center. Indian J Otolaryngol Head Neck Surg 2022;74(Suppl 3):6362-6. doi: 10.1007/s12070-021-02990-2.
- Funakoshi Y, Takeda S, Kadota Y, Maeda H. Mediastinal bronchogenic cyst with respiratory distress from airway and vascular compression. Thorac Cardiovasc Surg 2007;55:53-4. doi: 10.1055/s-2006-924002.
- Roland N, Bradley PJ. Neck swellings. BMJ 2014;348:g1078. doi: 10.1136/bmj.g1078.
- Nobuhara KK, Gorski YC, La Quaglia MP, Shamberger RC. Bronchogenic cysts and esophageal duplications: Common origins and treatment. J Pediatr Surg 1997;32:1408-13. doi: 10.1016/s0022-3468(97)90550-9.
- Abukhder M, Hulme J, Nathoo S, Shubhi S. Spontaneous retropharyngeal haematoma with direct oral anticoagulant medication. BMJ Case Rep 2021;14:e240369. doi: 10.1136/ bcr-2020-240369.
- Chan CC, Awobem A, Binns C, Jassar P. A rare case of spontaneous thyroid cyst haemorrhage. J Laryngol Otol 2012;126:960-2. doi: 10.1017/S0022215112001430.
- Ueda H, Baba H, Ondo K. Chronic expanding hematoma of thorax extended to the neck. Ann Thorac Cardiovasc Surg 2014;20 Suppl:535-8. doi: 10.5761/atcs.cr.12.02055.