

Suprasternal Bronchogenic Cyst in an 8-Year-Old Girl

Aseha Altun¹ , Giada Bilotta¹ , Gilles Van Haesendonck² , Callum Farris² , Olivier M Vanderveken^{2,3} , Anne Sieben⁴ , An Boudewyns^{2,3} 

¹Department of Translational Neurosciences, University of Antwerp Faculty of Medicine and Health sciences, Antwerp, Belgium

²Department of ENT, Head and Neck Surgery, Antwerp University Hospital, Antwerp, Belgium

³Department of Translational Neurosciences, University of Antwerp Faculty of Medicine and Health Sciences, Antwerp, Belgium

⁴Department of Pathology, Antwerp University Hospital, Antwerp, Belgium

Cite this article as: Altun A, Bilotta G, Van Haesendonck G, et al. Suprasternal bronchogenic cyst in an 8-year-old girl. B-ENT. 2023;19(4):245-248.

ABSTRACT

In this case report, we present an 8-year-old girl who was referred to the ear–nose–throat department of the Antwerp University Hospital because of an asymptomatic midline neck mass. After resection, histopathological examination was characteristic of a bronchogenic cyst. Bronchogenic cysts are rare, congenital anomalies of the tracheobronchial tree caused by aberrant bud formation of the tracheobronchial tree during embryogenic development. They usually have an intrathoracic location, whereas cervical, cutaneous, and subcutaneous cysts are extremely rare. Initial symptoms are respiratory distress, coughing, and bronchopulmonary infections by compression of the tracheobronchial tree. Imaging studies are recommended to determine the mass's tissue characteristics as well as the engagement of surrounding anatomical structures, which is necessary for adequate preoperative planning. However, a definitive diagnosis may be obtained only after surgery through histopathological analysis.

Keywords: Bronchogenic cyst, child, neck mass, asymptomatic, extrathoracic

Introduction

Bronchogenic cysts are uncommon congenital abnormalities of the tracheobronchial tree,¹ with an incidence of 1 in 42 000 to 68 000 individuals² and a male-to-female ratio of 4 : 1.²⁻⁷ They tend to occur more frequently in pediatric patients and are rare in adults.⁸

They arise during embryologic development due to an abnormal budding of the tracheobronchial tree. Although they typically have an intrathoracic location,^{8,9} extrathoracic presentations have been reported and can occur at the suprasternal notch, sternal manubrium, cervical region, tongue base, scapular area, shoulder, paravertebral, retroperitoneal, cutaneous, etc.³

Cutaneous, cervical, and subcutaneous cysts are very rare.^{10,11} The common sites of a subcutaneous bronchogenic cyst are the suprasternal region and the sternal manubrium.^{4,12}

Clinical presentation varies with patient age. Bronchogenic cysts are frequently symptomatic during infancy. Initial symptoms include shortness of breath, coughing, and bronchopulmonary infections due to compression of the

tracheobronchial tree.⁸ Subcutaneous bronchogenic cysts are discovered as an asymptomatic cervical mass or draining sinus shortly after birth or in early infancy.³ In this case report, we will discuss a rare case of a suprasternal bronchogenic cyst in an 8-year-old girl.

Case Presentation

An 8-year-old girl was referred by a pediatrician because of an asymptomatic midline neck mass. There was no change in volume over time and no signs of inflammation or pain. Clinical investigation revealed a soft, tender mass at the level of the jugular notch. The overlying skin was unaffected (Figure 1). Lymphadenopathy was absent. Further clinical examination of the ear–nose–throat region was unremarkable. The child had no medical history and was in good health condition.

Ultrasound of the mass showed a fluid-containing, cystic structure with a thin, regular wall on the midline, superior to the suprasternal notch. The cyst was located subcutaneously and measured 20 × 16 × 25 mm. There was no connection to the larynx or thyroid gland.

Corresponding Author: Gilles Van Haesendonck, e-mail: gilles.vanhaesendock@uza.be

Received: December 2, 2022 **Accepted:** June 27, 2023 **Publication Date:** October 26, 2023

Available online at www.b-ent.be



CC BY 4.0: Copyright@Author(s), "Content of this journal is licensed under a Creative Commons Attribution 4.0 International License."

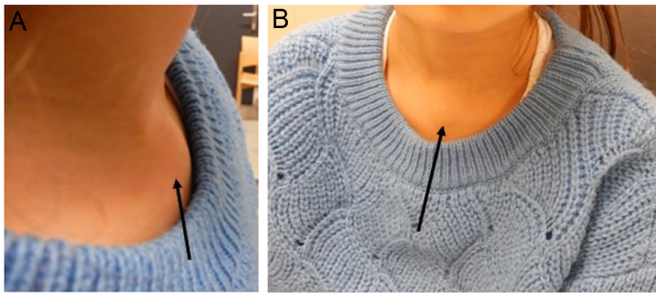


Figure 1. Clinical picture of the midline neck mass in lateral (A) and anterior (B) view.



Figure 2. Sagittal T2-weighted Dixon MRI showing a cystic structure in the subcutis with a homogeneous hyperintense signal. MRI, magnetic resonance imaging.

Additional examination with magnetic resonance imaging (MRI) confirmed the presence of a cystic structure in the subcutis, located superior to the suprasternal notch, and caudal to the thyroid isthmus. After gadolinium injection, a definite,

Main Points

- Bronchogenic cysts must be considered in the differential diagnosis of congenital cervical cysts.
- A definitive diagnosis of a congenital cyst can only be made by histopathological examination.
- Complete surgical excision of bronchogenic cysts is mandatory because of possible complications such as expansion, compression of surrounding structures, hemorrhage, infection, and the potential for developing malignancy.

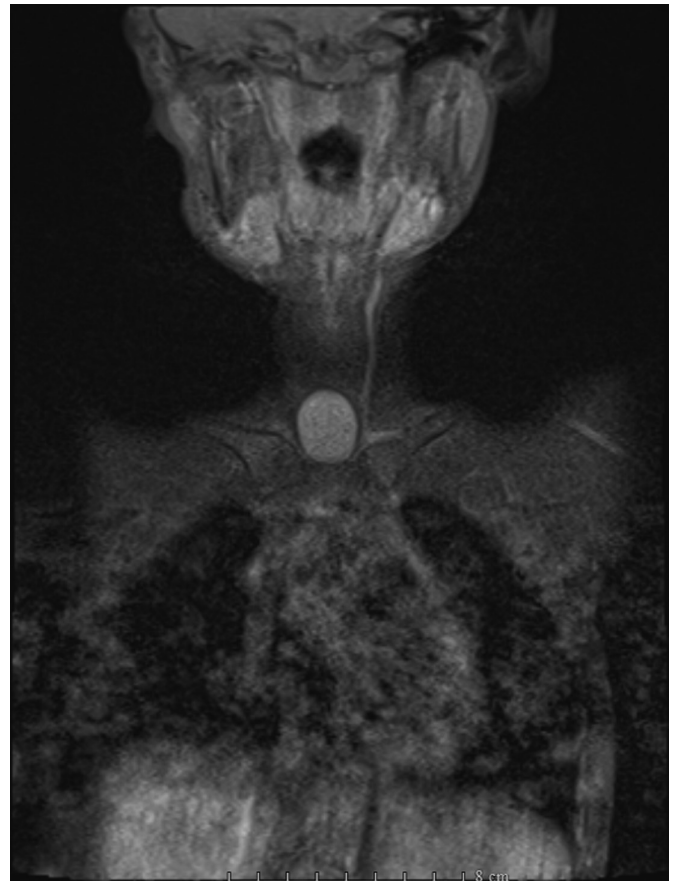


Figure 3. Coronal T1-weighted Dixon MRI of the cystic structure. MRI, magnetic resonance imaging.

homogeneous hyperintense signal was on T2-weighted MRI. These MRI findings were interpreted as a dermoid cyst (Figure 2-3).

Surgical resection of the cyst was performed under general anesthesia to allow a definitive histopathological diagnosis.

The surgical approach involved a horizontal neck incision superior to the cyst and blunt dissection on the cyst wall using bipolar forceps. The cyst could be totally removed, and the cyst wall remained intact. The postoperative course was uneventful.

The excised cyst measured 30 × 20 × 9 mm and showed no specific features. It was pink-colored and sharply margined with smooth borders. On section, the cyst was filled with a white, viscous substance. Microscopically the inner surface was lined with ciliated columnar epithelium containing a few mucous goblet cells without cytonuclear atypia. The cyst was filled with mucinous material which included some detached epithelial cells (Figure 4). These morphologic characteristics were compatible with a histopathological diagnosis of a bronchogenic cyst.

Discussion

In this case report, a bronchogenic mass presented as an asymptomatic midline neck mass in an 8-year-old girl. According to the literature, bronchogenic cysts are more common in males and are usually symptomatic during infancy.²⁻⁷

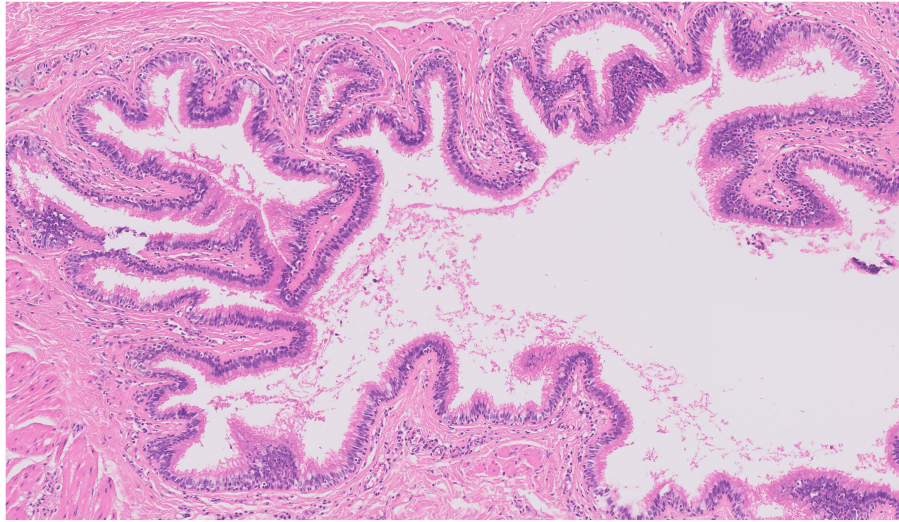


Figure 4. Microscopy: Hematoxylin-eosin stain showing the wall of the cyst lined with ciliated epithelium.

Extrathoracic bronchogenic cysts are generally present as an asymptomatic (anterior) cervical mass, like our case. Pressure symptoms such as dyspnea, cough, and dysphagia can occur with larger cysts. In contrast, intra-thoracic cysts are usually symptomatic.¹³

The differential diagnosis of congenital cervical cysts in children includes branchial cleft cysts, epidermal inclusion cysts, thymic cysts, thyroid cysts, dermoids and lymphangiomas, cystic teratomas, thyroglossal duct cysts, bronchogenic cysts, and cystic hygromas.^{1,2,12} The location of the cyst in the neck can be helpful during the diagnostic process.

Bronchogenic cysts appear in the fifth week of pregnancy. At that stage, the tracheobronchial tree consists of the anterior trachea and the posterior esophagus.² Instead of developing bronchial buds, that give rise to the respiratory tree, these cysts detach and migrate.² As the tracheobronchial tree evolves, this aberrant budding process continues.² Although they usually develop in or close to the tracheobronchial tree, bronchogenic cysts can also be found in other locations, such as the abdomen and neck.²

There are several imaging modalities that can guide the diagnosis of bronchogenic cysts. For adequate preoperative planning, it is also recommended to determine the mass's tissue characteristics as well as the engagement of surrounding anatomical structures.² On ultrasound, they appear as anechoic masses with or without hyperechoic debris. Ultrasound is particularly used in children, restricted to cysts with an acceptable image window.² On computerized tomography (CT), these cysts have a spherical shape with fine walls that are smooth or lobulated.² They are homogeneous, hypoattenuating, and show no contrast enhancement.² Some cysts show fluid levels and calcifications.² Bronchogenic cysts have a medium-to-high signal intensity on T1-weighted MRI images and a very high signal intensity on T2-weighted images.² This is because the cysts contain water as well as proteinaceous fluid.²

Although imaging is important, the final diagnoses can only be made after histopathological examination as shown in the

present case in which imaging studies suggested the presence of a dermoid cyst.^{2,8}

Surgical excision of bronchogenic cysts is mandatory because of possible complications such as expansion, compression of surrounding structures, hemorrhage, infection, and the potential for developing malignancy.^{3,6,9} The morbidity rate of surgical resection in the neck is low because the cyst can be easily separated from surrounding structures. However, the recurrent laryngeal nerve and large vessels can be at risk, depending on the location of the cyst.

Complete removal is important to avoid recurrence.^{5,8} Simple aspiration is therefore not an adequate treatment.¹³

Histopathological confirmation after surgery is required for the definitive diagnosis.⁹ These cysts are lined with respiratory epithelium, more specifically ciliated pseudostratified cylindrical epithelium covering a fibrous connective tissue wall with cartilaginous plates and seromucous subcutaneous glands.⁹ Smooth muscle cells may be found in this wall.⁹ The most valid diagnostic criterion is the appearance of cartilage plates in the wall of the cyst, with submucosal glands and smooth muscle cells.^{6,9,10,12}

Conclusion

Bronchogenic cysts arise during embryologic development due to an abnormal budding of the tracheobronchial tree. They are more frequently intra-thoracically located but can also be found extra-thoracically. The clinical presentation depends on age. Although these cysts are usually asymptomatic, they can also present with symptoms suggestive of respiratory distress, infection, or malignancy. Imaging modalities are used to guide the diagnoses and for preoperative planning but are not conclusive. The definitive diagnosis can only be made with histopathological confirmation after complete surgical resection of the cyst.

Informed Consent: Written informed consent was obtained from the patients' parents who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – A.A., G.B., A.S., A.B., G.V.H.; Design – A.A., G.B., A.B., G.V.H.; Supervision – A.B., G.V.H., O.V., C.F.; Resources – A.A., G.B.; Materials – G.B., A.S.; Data Collection and/or Processing – A.A., G.B., A.S.; Analysis and/or Interpretation – A.A., G.B., A.S., G.V.H., O.V.; Literature Search – A.A., G.B., G.V.H.; Writing – A.A., G.B., A.S., A.B., C.F.; Critical Review – A.B., G.V.H., O.V., C.F.

Declaration of Interests: The authors have no conflict of interest to declare.

Funding: The authors declared that this study has received no financial support.

References

1. Shah SK, Stayer SE, Hicks MJ, Brandt ML. Suprasternal bronchogenic cyst. *J Pediatr Surg*. 2008;43(11):2115-2117. [\[CrossRef\]](#)
2. Cohn JE, Rethy K, Prasad R, Mae Pascasio J, Annunzio K, Zwillenberg S. Pediatric bronchogenic cysts: a case series of six patients highlighting diagnosis and management. *J Invest Surg*. 2020;33(6):568-573. [\[CrossRef\]](#)
3. Pujary K, Pujary P, Shetty R, Hazarika P, Rao L. Congenital cervical bronchogenic cyst. *Int J Pediatr Otorhinolaryngol*. 2001;57(2):145-148. [\[CrossRef\]](#)
4. Ramón R, Betloch I, Guijarro J, Bañuls J, Alfonso R, Silvestre JF. Bronchogenic cyst presenting as a nodular lesion. *Pediatr Dermatol*. 1999;16(4):285-287. [\[CrossRef\]](#)
5. Manchanda V, Mohta A, Khurana N, Das S. Subcutaneous bronchogenic cyst. *J Cutan Aesthet Surg*. 2010;3(3):181-183. [\[CrossRef\]](#)
6. Gaikwad P, Muthusami JC, Raj JP, Rajinikanth J, John GM. Subcutaneous bronchogenic cyst. *Otolaryngol Head Neck Surg*. 2006;135(6):951-952. [\[CrossRef\]](#)
7. Zvulunov A, Amichai B, Grunwald MH, Avinoach I, Halevy S. Cutaneous bronchogenic cyst: delineation of a poorly recognized lesion. *Pediatr Dermatol*. 1998;15(4):277-281. [\[CrossRef\]](#)
8. Bocciolini C, Dall'olio D, Cunsolo E, Latini G, Gradoni P, Laudadio P. Cervical bronchogenic cyst: asymptomatic neck mass in an adult male. *Acta Otolaryngol*. 2006;126(5):553-556. [\[CrossRef\]](#)
9. Ustundag E, Iseri M, Keskin G, Yayla B, Muezzinoglu B. Cervical bronchogenic cysts in head and neck region. *J Laryngol Otol*. 2005;119(6):419-423. [\[CrossRef\]](#)
10. Mangiameli G, Arame A, Le Pimpec-Barthes F. Giant presternal subcutaneous bronchogenic cyst: a rare but possible occurrence. *Asian Cardiovasc Thorac Ann*. 2020;28(8):507-509. [\[CrossRef\]](#)
11. Lee HS, Park CK, Joo KB, et al. Subcutaneous bronchogenic cyst: unusual ultrasonographic findings. *J Ultrasound Med*. 2001;20(5):563-566. [\[CrossRef\]](#)
12. Moon SM, Lee SM, Kang H, Choi HJ. Presternal subcutaneous bronchogenic cyst in adolescence: a case report and unusual ultrasonographic findings. *Med (Baltim)*. 2017;96(5):e6054. [\[CrossRef\]](#)
13. Mehta RP, Faquin WC, Cunningham MJ. Cervical bronchogenic cysts: a consideration in the differential diagnosis of pediatric cervical cystic masses. *Int J Pediatr Otorhinolaryngol*. 2004;68(5):563-568. [\[CrossRef\]](#)