CASE REPORT

Bronchogenic cyst—a rare case mimicking a laryngocoele

Mohammed Farid1,* and Philip Michael2

1Head and Neck Surgery, The Royal London Hospital, Barts Health NHS Trust, London, UK and 2Department of Otolaryngology, Royal Victorian Eye and Ear Hospital, Melbourne, Australia

*Correspondence address. E-mail: mohammed.farid@gmail.com

Abstract

Midline neck lumps are commonly recognized in clinical practice. Nonetheless, we present an unusual case of a 24-year-old Bangladeshi man who has a slowly growing midline neck lump. The characteristic lump did not move on swallowing and was more pronounced on coughing suggesting an external laryngocoele as a main differential. The patient underwent urgent surgical excision of the lump. Further histopathology findings were consistent with the diagnosis of a bronchogenic cyst which is deemed rare with such presentation.

INTRODUCTION

Bronchogenic cysts are rare congenital malformations arising from the tracheobronchial tree that rarely present outside the thorax. The unusual sign of lump expansion with increased intrathoracic pressure (Valsalva manoeuvre) was indicative of an airway communication, though this was not evident on imaging (Computed tomography, CT).

The pathophysiology of bronchogenic cysts illustrated the diagnostic difficulties encountered with rare lesions and highlighted the awareness of the different anatomical presentations.

CASE REPORT

A 24-year-old unemployed man of Bangladeshi origin presented with a 12-month history of a slowly growing midline neck swelling. There were no associated aero-digestive symptoms, relevant occupational or social history.

Clinical examination revealed a 6 cm diameter midline, soft, fluctuant, non-tender mass, which did not move on swallowing. However, the swelling was seen to become more prominent when performing a Valsalva manoeuvre (Figs 1 and 2). The remainder of the otolaryngology examination including flexible naso-laryngendoscopy was unremarkable. Owing to the lump enlargement with increased intrathoracic pressure, an initial differential diagnosis of a laryngocoele was made.

CT scan (Fig. 3) demonstrated a well-defined 5 × 4 cm cystic lesion with no clear communication to the larynx. Aspirate of the cyst appeared mucoid and did not culture any microorganisms.

The specimen was completely excised through a low-neck skin crease incision. It had a pedicle onto the superior border of the sternal notch with a portion of intervening cartilage. A 5 × 4 × 4 cm cystic lesion containing mucoid fluid was resected (Fig. 4). Histopathology report confirmed that the cyst contained a portion of intervening cartilage with a pseudostratified columnar epithelium lining. The origin from the tracheobronchial tree added to the diagnostic certainty of bronchogenic cyst.

DISCUSSION

Bronchogenic cysts, also known as foregut duplication cysts, are uncommon, benign, congenital lesions representing accessory bronchial buds. These cysts are found predominately in...
men (M:F ratio 4:1) with a prevalence of 1 in 42 000 to 1 in
68 000 [1, 2].

The cysts arise due to an abnormal budding of the ventral
foregut (the future tracheobronchial tree) during lungs develop-
ment. If these abnormal buds are resorbed, no further clinical
sequelae arise. However, persistent buds may migrate during
the course of development and rest in intrathoracic or extra-
thoracic region. The intrathoracic location is the mediastinum,
pericardium, diaphragm and lungs while the extrathoracic site
is the abdomen, oesophagus, the retroperitoneum, the subcuta-
neous tissue or the neck. In terms of the head and neck region,
cysts can be in the suprasternal notch, pre sternum, shoulder,
infraoral, infraclavicular region, posterior pharyngeal wall or
extend into the mediastinum. A superficial pre sternal or
suprasternal location is most common, while deep or lateral
neck bronchogenic cysts are rare. Intra dural extramedullary
bronchogenic cysts in the high and mid cervical region are
reported. Bronchogenic cysts are classified by Maier according
to their site of origin into paratracheal, carinal, hilar, paraoeso-
phageal, and atypical such as diaphragmatic, abdominal, intra-
cutaneous, subcutaneous, or subcapular [3–7].

The course of upward migration begins around the carina
and extends upwards either parallel to the trachea deep into
the neck or towards the skin superficially. Superficial migra-
tion, similar to our case, is explained by tracheal bud develop-
ment around the fifth week of development when the right and
left mesenchymal bars displace the cyst anteriorly prior to their
fusion. Consequently, abnormal budding during the develop-
ment of the trachea may result in a midline cervical cyst
whereas a lateral neck cyst may form during the development
of the bronchial system. The localization of the cyst in the cer-
vical region therefore suggests that the anomaly occurred
before the fifth week of gestation. About 70% of more than the
total 70 reported cases of cervical bronchogenic cysts are in the
midline region. Bronchogenic cysts of the lateral neck are
located in lower cervical regions whereas the upper cervical
cysts are generally on the midline [8–11].

The histological finding of respiratory epithelium alone
would not be sufficient to diagnose bronchogenic cysts as the
same may be expected in laryngocoeles. The most reliable cri-
terion for the diagnosis of bronchogenic cysts has been the
presence of cartilage plates in the wall of the cyst, with sub-
mucosal glands and smooth muscles [12, 13].

Early surgical intervention can prevent future symptoms and
complications, which may make subsequent resection more
challenging. If left untreated, bronchogenic cysts may undergo
squamous metaplasia, inflammation, or mucosal necrosis and
thus complicate diagnostic confirmation. Furthermore, deep
cysts may become infected and present as a neck abscess. The
importance of resecting extrathoracic superficial bronchogenic

Figure 1: The appearance of suprasternal midline neck lump pre Valsalva
manoeuvre.

Figure 2: The midline neck lump is more prominent at Valsalva manoeuvre.

Figure 3: Cross sectional image of midline neck lump with no tracheal
communication.

Figure 4: Intra operative image of bronchogenic cyst prior to surgical excision.
cysts has been highlighted in a number of cases, reporting mucoepidermoid carcinoma or recurrent malignant melanoma transformation as potential complications [14–18].

The differential spectrum of a midline neck mass include thyroglossal cysts, branchial cleft cysts or thymic cysts. Unlike branchial cysts or thyroglossal cysts, bronchogenic cysts can track towards the sternal notch or have a cranial extension. Other differentials for neck cysts include cystic lymatatic, lipomas, thyroid cysts, teratomas, epidermoid cysts and cystic norinomas. The clinical presentation and histological nature of these cysts is distinct from bronchogenic cysts [19, 20].

The expansile nature of the presented rare midline neck lump suggested a top differential diagnosis of an external laryngocoele due to the distinguished typical presentation superior to the laryngeal cartilage after passing through the thyrohyoid membrane. While bronchogenic cysts have not previously been described as mimicking laryngocoeles, a previous case of a lipoma presenting as a midline neck lump with shape change during valsalva manoeuvre was reported previously [21].

The pre-operative diagnosis of bronchogenic cysts based on history and imaging were only accurately made in a third of intrathoracic cases [22]. This case of bronchogenic cyst was diagnosed based on combined histological, clinical and radiological findings.

CONFICT OF INTEREST STATEMENT

None declared.

REFERENCES

1. Gaikwad P, Muthusami JC, Raj JP, Rajinikanth J, John GM. Subcutaneous bronchogenic cyst. Otolaryngol Head Neck Surg 2006;135:951–2.
2. Coselli MP, de Ipolyi P, Bloss RS, Diaz RF, Fitzgerald JB. Bronchogenic cysts above and below the diaphragm: report of eight cases. Ann Thorac Surg 1987;44:491–4.
3. Maier HC. Bronchogenic cysts of the mediastinum. Ann Surg 1948;127:476–502.
4. Moz U, Gamba P, Pignatelli U, D’Addazio G, Zorzi F, Fiaccavento S, et al. Bronchogenic cysts of the neck: a rare localization and review of the literature. Acta Otorhinolaryngol Ital 2009;29:36–40.
5. Pujaory K, Pujaory P, Shetty R, Hazarika P, Rao L. Congenital cervical bronchogenic cyst. Int J Pediatr Otorhinolaryngol 2001;57:145–8.
6. Yerman HM, Holinger LD. Bronchogenic cyst with tracheal involvement. Ann Otol Rhinol Laryngol 1990;99:89–93.
7. Lee DH, Yoon TM, Lee JK, Lim SC. Bronchogenic Cyst in the Head and Neck Region. J Craniofac Surg 2017; [Epub ahead of print].
8. Swanson SJ III, Skoog SJ, Garcia V, Wahl RC. Pseudoadrenal mass: unusual presentation of bronchogenic cyst. J Pediatr Surg 1991;26:1401–3.
9. Halsleton PS. Spencer’s Pathology of the Lung. New York: McGraw-Hill, 1996;1283.
10. Ustundag E, Iseri M, Keskin G, Yayla B, Muezzinoğlu B. Cervical bronchogenic cysts in head and neck region. J Laryngol Otol 2005;119:419–23.
11. 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;126:553–6.
12. Sedwick JD, Giannoni C. Bronchogenic cyst of the oropharynx and hypopharynx in a neonate. Otolaryngol Head Neck Surg 2001;125:105–6.
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;68:563–8.
14. Houser WC, Dorff GJ, Rosenzweig DY, Aussem JW. Mycobacterial infection of a congenital bronchogenic cyst. Thorax 1980;35:312–3.
15. McManus K, Holt GR, Audeforte TM, Trinkle JK. Bronchogenic cyst presenting as deep neck abscess. Otolaryngol Head Neck Surg 1984;92:109–14.
16. Hadjiyanassas E, Ray J, Ryhs-Williams S. A cervical bronchogenic cyst in an adult male. Eur Arch Otorhinolaryngol 2003;260:216–8.
17. Tanita M, Kikuchi-Numagami K, Ogoshi K, Suzuki T, Tabata N, Kudoh K, et al. Malignant melanoma arising from cutaneous bronchogenic cyst of the scapular area. J Am Acad Dermatol 2002;46:S19–21.
18. Tanaka M, Shimokawa R, Matsubara O, Aoki N, Kaminaya R, Kasuga T, et al. Mucoepidermoid carcinoma of the thymic region. Acta Pathol Jpn 1982;32:703–12.
19. Dolgin SE, Groisman GM, Shah K. Subcutaneous bronchogenic cysts and sinuses. Otolaryngol Head Neck Surg 1995;112:763–6.
20. Rapado F, Bennett JD, Stringfellow JM. Bronchogenic cyst: an unusual cause of lump in the neck. J Laryngol Otol 1998;112:893–4.
21. Ramakantan R, Shah P. Anterior neck lipoma masquerading as an external laryngocoele. J Laryngol Otol 1989;103:1087–8.
22. Shimazu R, Kuratomy Y, Inokuchi A. A case of an upper cervical bronchogenic cyst in an adult. Auris Nasus Larynx 2006;33:351–3.