The Journal of Laryngology & Otology (2010), ***124***, *1325–1328.*

doi:10.1017/S0022215110000423

Clinical Record

Recurrent neck abscess due to a bronchogenic cyst in an adult

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Abstract

Background: Neck abscesses can originate from congenital cervical cysts. Cervical cysts of bronchogenic origin are rare and often asymptomatic. Common symptoms of bronchogenic cysts are stridor, dyspnoea and dysphagia. The reported patient represents the second published case of a bronchogenic cyst causing a neck abscess in an adult.

Case report: We report a case of a cervical bronchogenic cyst presenting as a recurrent supraclavicular abscess in a middle-aged woman. During extirpation, a fistula was demonstrated to the right upper lobe of the lung, suspected because the cyst inflated synchronously with respiration.

Discussion: The symptoms of bronchogenic cysts are due to the effects of compression or fistulas. In the majority of these cysts, a thorough investigation involving history, examination and radiological imaging does not clearly demonstrate a fistula. Therefore, extirpation is both diagnostic and therapeutic.

Conclusion: A bronchogenic cyst is a very rare cause of a recurrent deep neck abscess. Total extirpation is the treatment of choice.

Key words: Bronchogenic Cyst; Neck Abscess; Adult

Introduction

Acute, severe neck abscesses are commonly encountered. They can be caused by infections originating from the head, by primary or secondary infection of the cervical lymph nodes or thyroid gland, or by infected congenital cysts.

Congenital cervical cysts are common in adults, and are often derived from the branchiogenic cleft, thymus or thyroglossal duct. Cervical cysts of bronchogenic origin are rare.[1,2]

A bronchogenic cyst is a malformation of the ventral foregut. The cyst is located in the thorax in more than 50 per cent of cases.[3] Other locations include subcutaneous,[4,5] subscapular,[5] abdominal[6] and retroperitoneal[7] sites. Cervically located bronchogenic cysts are often asymptomatic,[1,8–12] and are generally detected in childhood.[1,10,13–15]

The symptoms caused by cervical bronchogenic cysts include stridor in neonates,[13–15] dyspnoea on exertion,[8,16] dysphagia[8,17] and recurrent infection.[17] One case report described a deep neck abscess due to a bronchogenic cyst.[18]

We describe a second case of a bronchogenic cyst presenting as a recurrent neck abscess in a middle-aged woman.

Case report

A 51-year-old woman was referred to our hospital for evaluation of a painful cervical swelling plus dysphagia of five days' duration. She had not noticed any fever or dyspnoea. The swelling had been present for as long as she could remember. Four years before admission, thyroid function tests had been performed, with normal results. She was also known to have Crohn's disease, and had bilateral hearing loss due to multiple attacks of sudden sensorineural hearing loss.

Physical examination showed a painful, tense mass located in the right supraclavicular and paratracheal area. Hypertrophic tonsils were seen in the oropharynx.

Computed tomography (CT) revealed a 4.5 cm diameter cyst with an air–fluid level located dorsal to the right thyroid lobe. The hypopharynx was compressed. No connections were seen with the lungs, trachea or thyroid gland. The most likely diagnosis was a cervical abscess, which was confirmed by surgical drainage. Rigid endoscopy of the oesophagus and the upper and lower airways did not locate any fistula. Bacterial culture of the abscess contents grew a mixture of different bacteria, but no specific pathogen was detected.

Similar complaints and signs recurred after three months. A second CT scan again showed a cyst with an air–fluid level, highly suspicious for a retrotracheal abscess. A gastrografin contrast swallow study demonstrated lateralisation of both the trachea and the oesophagus to the left. The abscess was drained surgically and a biopsy taken. Again, rigid endoscopy of the oesophagus and the upper and lower airways located no fistula. Histopathological examination of the biopsy identified pus and inflamed, ciliated, pseudo-stratified, columnar epithelium.

Three months later, when the patient had recovered completely and was free of symptoms, magnetic resonance

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Presented at the 7th annual meeting of the North German Fellowship of Otorhinolaryngology and Head and Neck Surgery, 1 April 2007, Kiel, Germany.
Accepted for publication: 4 January 2010. First published online 16 March 2010.

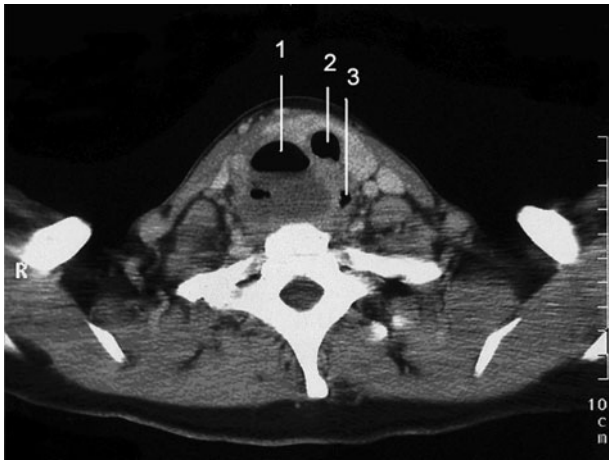

FIG. 1

Axial computed tomography scan of the neck, showing the bronchogenic cyst filled with fluid and air (1), and displacing the trachea (2) and oesophagus (3) to the left. R = right

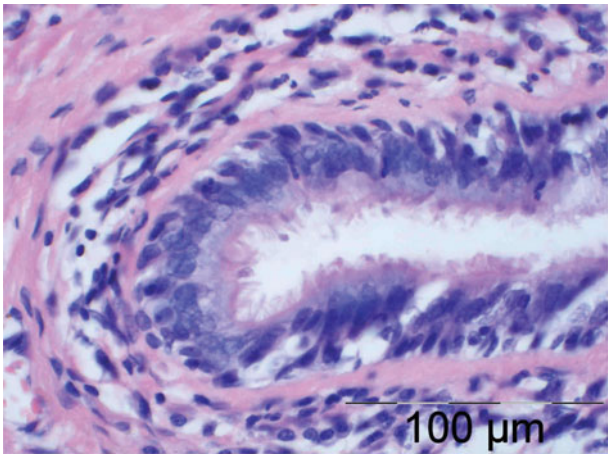

FIG. 3

Photomicrograph of a section of the cyst wall, showing ciliated, pseudostratified, columnar epithelium. Note that a submucosal layer with cartilage and smooth muscle is absent. (H&E; ×400)

imaging (MRI) and CT scanning of the cyst were performed (Figure 1). The cyst appeared to be filled with air, and a connection with the trachea was suspected. Ultrasonography revealed that the cyst filled with air during the Valsalva manoeuvre.

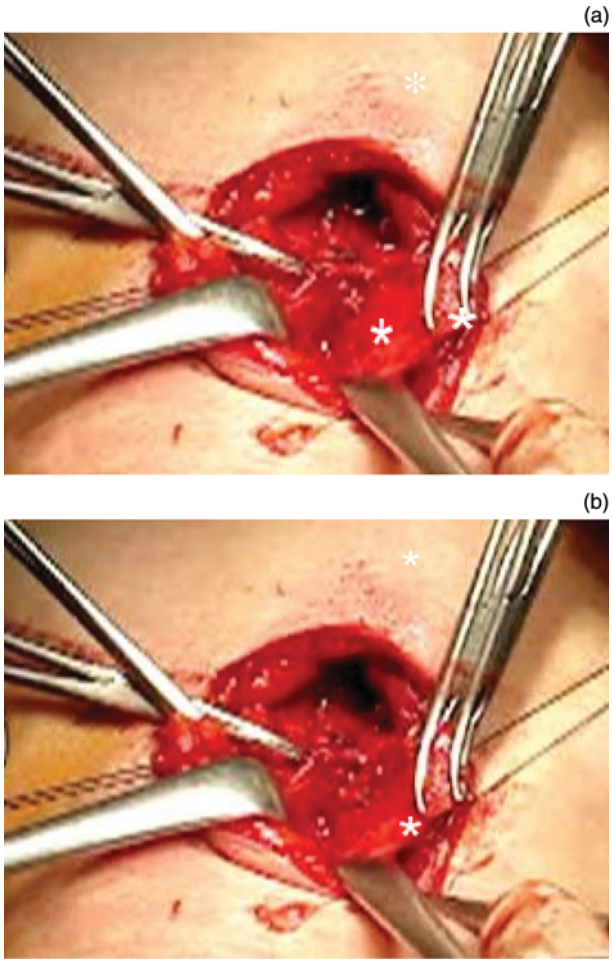

FIG. 2

Intra-operative photographs taken during extirpation, showing the cyst (*) inflated and deflated synchronous with (a) inspiration and (b) expiration, during artificial ventilation.

It was decided to again undertake surgery. Pre-operatively, methylene blue dye was injected into the cyst. Rigid endoscopy of the upper and lower airways again showed no fistula. The cyst was excised by a collar incision, and a thorough exposition and inspection was made of the trachea, sternocleidomastoid muscle, common carotid artery, external jugular vein, and vagal and recurrent nerves. The recurrent nerve was identified and bluntly separated from the ballooning cyst. The transparent, thin-walled cyst was seen to fill and empty synchronous with respiration (Figure 2). Because the endotracheal tube cuff pressed tightly against the trachea, a fistula had to be present somewhere between the lower airways (under the cuff) and the cyst. Upon opening of the cyst, no methylene blue dye was retrieved. Palpation of the entire trachea did not reveal any sign of a fistula. However, a strand of connective tissue connected to the upper right lobe was located and assumed to be the fistula, and was ligated. Subsequently, no air leak was seen while flooding the wound with water.

Histopathological examination of the cyst revealed ciliated, pseudo-stratified, columnar epithelium (Figure 3). No cartilage, smooth muscle or mucous glands were seen.

Post-operatively, the patient suffered a recurrent nerve palsy on the operated site, as detected by indirect laryngoscopy.

After three years' follow up, the patient was free of symptoms. Although her recurrent nerve palsy persisted, her voice improved with speech therapy.

Discussion

The symptoms of bronchogenic cysts are generally due to direct compression of surrounding structures, resulting in stridor, dyspnoea on exertion or dysphagia, or to a fistula resulting in recurrent infections or abscesses. During the Valsalva manoeuvre, bronchogenic cysts can increase in volume due to the presence of a fistula,[17] as in our case, or due to extension from an intrathoracic location into the supraclavicular area.[16] Most such fistulas are rarely[17] if ever[12] seen on CT or MRI scanning, and are not detected until surgical extirpation. Therefore, extirpation is both diagnostic and therapeutic. A fistula should be considered in symptomatic cases, and should be searched for during surgery.

There are two embryological theories about the origin of cervical bronchogenic cysts. In the fourth embryonic week,

https://doi.org/10.1017/S0022215110000423 Published online by Cambridge University Press

the laryngotracheal bud sprouts as the ventral appendix of the foregut, and forms the origin of the larynx and tracheobronchial bud.[19] The first theory holds that bronchogenic cysts derive from abnormal buddings of the distal part of the tracheobronchial bud. An alternative theory[20] hypothesises that bronchogenic cysts originate from preformed cysts of the thoracic cavity. During the development of the tracheobronchial tree and closure of the thoracic wall, this cyst pinches off and migrates to the neck region. In both theories, a connection to the thorax may persist as a fistula,[17] stalk[15] or connective tissue strand[20] to the mediastinum.

Diagnostic investigation includes a thorough history for cough, recurrent airway infection, dysphagia and dyspnoea. Physical and ENT examination should include the Valsalva manoeuvre[16,17] and transillumination of the cyst.[11] Radiological imaging of the cyst should comprise ultrasonography, gastrografin contrast swallow study, and CT and MRI scanning. Al-Kasspooles *et al.*[17] have demonstrated a fistula to the trachea using virtual bronchoscopy. Eventually, the diagnosis is made by means of rigid endoscopy (i.e. bronchoscopy, oesophagoscopy and laryngoscopy) and total extirpation of the cyst.

The differential diagnosis of a cervical neck cyst is extensive. It comprises branchial cysts, thyroglossal cysts, thymic cysts, thyroid cysts, lipomas, epidermal inclusion cysts, teratomas and cystic lymphoid vascular malformations. The location (i.e. medial or lateral) is not conclusive for the diagnosis, as has been suggested in the past.[13] Branchial cysts are a commoner, better known cause of neck abscesses. Branchial cysts, being of ectodermal origin, are usually characterised by stratified, squamous epithelium, and only rarely by pseudo-stratified, columnar, ciliated epithelium.[4,10] Theoretically, Crohn's disease may have caused an upper digestive tract fistula in our patient.[21] However, her Crohn's disease had been quiescent for many years, and no ulceration of the oral cavity or oesophagus was reported. Furthermore, thorough endosopic inspection did not reveal any ulceration or fistula. This possibility was therefore excluded.

- **Cervical bronchogenic cysts seldom cause symptoms after childhood**
- **This paper describes a neck abscess caused by a bronchogenic cyst in an adult**
- **The treatment of choice in this situation is total extirpation of the cyst, and its possible coexisting fistula, to avoid recurrent infection and malignant transformation**

Because the diagnosis of bronchogenic cyst is made histopathologically, there is an ongoing debate regarding strict diagnostic criteria. The structures of the bronchogenic cyst are embryologically derived from endoderm, with accessory tissue of mesodermal origin. The histological criteria for a bronchogenic cyst are therefore a cyst with ciliated, pseudo-stratified, columnar epithelium and seromucinous glands, cartilage and smooth muscle. In this way the histological appearance ranges from an alveolus to a small bronchus. Although these criteria are often met in the case of intrathoracic bronchogenic cysts,[3] in cervical bronchogenic cysts frequently only ciliated, pseudo-stratified, columnar epithelium is found.[9,15,17] Some authors refer to such cysts as foregut cysts,[6] and consider cartilage obligatory for the diagnosis of a bronchogenic cyst.[10] Others state that cartilage is seldom present.[15] In our case, however, the diagnosis was made at surgery, with the histopathological appearance supplying only supportive evidence.

The treatment of choice for bronchogenic cysts, irrespective of location or symptoms, is total extirpation, because of the risk of complications (e.g. abscess) and possible malignant transformation. Malignancies have been found in intrathoracic[3,7] and retroperitoneal[7] bronchogenic cysts, and are known to occur in asymptomatic branchial cysts.[22]

In our patient, recurrent nerve palsy was seen postoperatively. This complication has also been reported by McManus *et al.*[18] Despite such a serious complication, we still support the practice of extirpation of all cervical cysts, even asymptomatic ones.

References

1 Ustundag E, Iseri M, Keskin G, Yayla B, Muezzinoglu B. Cervical bronchogenic cysts in head and neck region: review of the literature. *J Laryngol Otol* 2005;**119**:419–23
2 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
3 Cuypers P, De Leyn P, Cappelle L, Verougstraete L, Demedts M, Deneffe G. Bronchogenic cysts: a review of 20 cases. *Eur J Cardiothorac Surg* 1996;**10**:393–6
4 Zvulunov A, Amichai B, Grunwald MH, Avinoach I, Halevy S. Cutaneous bronchogenic cyst: delineation of a poorly recognized lesion. *Pediatr Dermatol* 1998;**15**:277–81
5 Ozel SK, Kazez A, Koseogullari AA, Akpolat N. Scapular bronchogenic cysts in children: case report and review of the literature. *Pediatr Surg Int* 2005;**21**:843–5
6 Liang MK, Yee HT, Song JW, Marks JL. Subdiaphragmatic bronchogenic cysts: a comprehensive review of the literature. *Am Surg* 2005;**71**:1034–41
7 Sullivan SM, Okada S, Kudo M, Ebihara Y. A retroperitoneal bronchogenic cyst with malignant change. *Pathol Int* 1999;**49**:338–41
8 Newkirk KA, Krowiak EJ, Tassler AB, Deeb Z. Bronchogenic cysts of the neck in adults. *Ann Otol Rhinol Laryngol* 2004;**113**:691–5
9 Shimazu R, Kuratomi Y, Inokuchi A. A case of an upper cervical bronchogenic cyst in an adult. *Auris Nasus Larynx* 2006;**3**:351–3
10 Hadi UM, Jammal HN, Hamdan ALM, Saad AM, Zaatari GZ. Lateral cervical bronchogenic cyst: an unusual cause of a lump in the neck. *Head Neck* 2001;**23**:590–3
11 Gaikwad P, Muthusami JC, Raj JP, Rajinikanth J, John GM. Subcutaneous bronchogenic cyst. *Otolaryngol Head Neck Surg* 2006;**135**:951–2
12 Eng CY, Quraishi MS, Watson MG. Bronchial or branchial cervical cyst? *Otolaryngol Head Neck Surg* 2006;**135**:958–9
13 Touloukian RJ. Air filled bronchogenic cyst presenting as a cervical mass in the newborn. *J Pediatr Surg* 1982;**17**:311–12
14 Lai P, Nguyen LH, Kim PC, Campisi P. An unusual case of biphasic stridor in an infant: suprasternal bronchogenic cyst. *J Pediatr* 2006;**149**:424
15 Maung KH, Low C, Knight LC, Cullinane CJ. Multiple cervical bronchogenic cysts. *J Laryngol Otol* 2006;**120**:145–7
16 Park OK, Buford CH. Bronchogenic cyst of neck and superior mediastinum. *Ann Surg* 1955;**142**:130–3
17 Al-kasspooles MF, Alberico RA, Douglas WG, Litwin AM, Wiseman SM, Rigual NR *et al*. Bronchogenic cyst presenting as a symptomatic neck mass in an adult: case report and review of the literature. *Laryngoscope* 2004;**114**:2214–17
18 McManus K, Holt GR, Aufdemorte TM, Trinkle JK. Bronchogenic cyst presenting as deep neck abscess. *Otolaryngol Head Neck Surg* 1984;**92**:109–14

https://doi.org/10.1017/S0022215110000423 Published online by Cambridge University Press

19 Sadler TW. Respiratory system. In: Sadler TW, ed. *Langman's Medical Embryology*, 7th edn. Baltimore: Williams & Wilkins, 1995;232–41
20 Pujary K, Pujary P, Shetty R, Hazarika P, Rao L. Congenital cervical bronchogenic cyst. *Int J Pediatr Otorhinolaryngol* 2001;**57**:145–8
21 Mills CC, Amin M, Manisali M. Salivary duct fistula and recurrent buccal space infections: a complication of Crohn's disease. *J Oral Maxillofac Surg* 2003;**61**:1485–7
22 Lanzafame S, Caltabiano R, Puzzo L, Cappellani A. Thyroid transcription factor 1 (TTF-1) and p63 expression in two primary thyroid papillary carcinomas of branchial cleft cysts. *Virchows Arch* 2006;**449**:129–33

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Dr A J C Hazenberg takes responsibility for the integrity of the content of the paper.
Competing interests: None declared

https://doi.org/10.1017/S0022215110000423 Published online by Cambridge University Press