Case Reports

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Intrathyroidal Bronchogenic Cysts: A report of two cases

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Abstract

Bronchogenic cysts are rare congenital anomalies arising from an abnormal budding of the primitive foregut or tracheobronchial tree. They are most commonly identified in the mediastinum though they can be found in the lung. Ectopic bronchogenic cysts are uncommon in clinical practice, and even rarer when located in the thyroid gland. We report here two cases of intrathyroidal bronchogenic cysts and discuss the patients' outcomes.

Keywords

Ectopic, thyroid, bronchogenic cyst, complications

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Introduction

Bronchogenic cysts are rare congenital cystic lesions that are thought to arise due to dysplasia in the foregut during embryogenesis.[1] In adults, the most common location of the cysts is the thorax, either the mediastinum or the lung parenchyma.[1] However, bronchogenic cysts have been reported in several extra-thoracic locations.[1–4] The cysts are thought to arise from abnormal budding of the primitive foregut which gives rise to the respiratory tract and the oesophagus and explains why the majority of the cysts are in the chest. Most cysts are spherical, singular, unilocular, fluid-filled and are not in communication with the tracheobronchial system.[3,5] Bronchogenic cysts are mostly asymptomatic unless the size is large enough to compress surrounding tissues or when secondary infection occurs.[6,7]

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We report here two cases of intrathyroidal bronchogenic cysts, and discuss some related issues and relevant literature.

Case reports

Case 1

A 26-year-old woman was admitted to hospital one month after a left thyroid nodule was discovered during a physical examination by her local physician. The patient had no pain, palpitations, tremor, hoarseness, or hypocalcaemia-induced convulsions. No palpable nodules or lymph nodes in the thyroid gland or neck were observed during a physical examination upon hospital admission. An ultrasound examination was performed, which showed a left thyroid nodule (9×5 mm) with hypo-echogenicity, unclear margins, variable echogenicity, and microcalcifications (Figure 1a); it was classified as Thyroid Imaging Reporting and Data System (TIRADS) stage 4c (i.e., highly suspicious nodules [50–85% risk of malignancy]).[8] No swollen lymph nodes were found in the patient's neck and no nodule was observed in the right thyroid. Colour Doppler flow imaging (CDFI) showed an area of hypo-echogenicity with a small blood flow signal. A neck computed tomography (CT) scan showed a low-density nodule (9×6 mm) in the dorsal left thyroid, within which small air bubbles were seen (Figure 1b). The nodule was considered to be a neoplastic lesion. The patient's thyroid function, thyroid autoantibodies, and calcitonin were within normal ranges.

Fine needle aspiration biopsy (FNAB) was not performed because the nodule

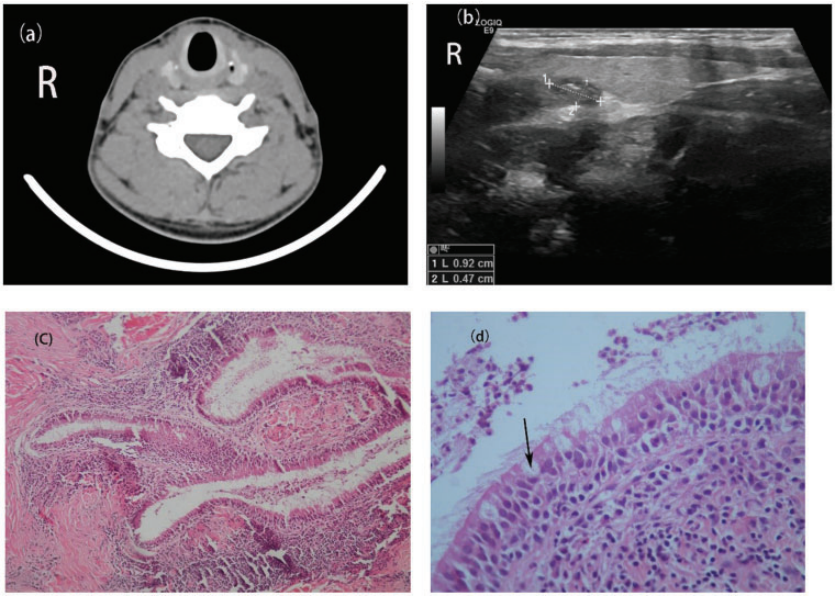

Figure 1. (a) A computed axial tomography (CAT) scan of the neck without contrast, showing a bubble in the mass encapsulated by the thyroid. (b) Thyroid ultrasound showed one solid and hypoechoic nodule of 9×5 mm in the left thyroid gland, with heterogeneous internal echoes and hyperechoic spots. (c) Postoperative pathology findings showed that the cyst wall was lined by a pseudostratified ciliated columnar epithelium, with fibrous connective tissue proliferation and lymphocytic infiltration. (Haematoxylin and eosin [H&E] stain, 100× magnification). (d) The histopathological section magnified at 400× shows the goblet cells (black arrow) among the epithelial cells.

was located in a crucial site where the recurrent laryngeal nerve enters the larynx. After preoperative preparation, the patient underwent left thyroid gland lobectomy under general anaesthesia. Intraoperative exploration showed a palpable pliable nodule in the dorsal middle-upper part of the left thyroid, approximately 1.0 ×1.0 cm in size. The membrane of the gland had not been invaded. The intraoperative frozen section histological examination showed that the nodule in the left thyroid was a benign lesion. Because the lesion was classified as TIRADS stage 4c, left central lymph node dissection was performed and a drainage tube was inserted. From postoperative pathological examination of the tissues, a cyst was identified in the thyroid tissue and the cyst wall was lined by a pseudostratified ciliated columnar epithelium, with proliferation of fibrous connective tissue and an infiltration of lymphocytes (Figure 1c). Goblet cells were also seen (Figure 1d). The left central lymph nodes showed reactive hyperplasia. The patient was diagnosed as having an intrathyroidal ectopic bronchogenic cyst.

The drainage tube was removed on Day 3 post-surgery, and on Day 4, the patient was discharged from hospital. Three days later, the patient was readmitted to hospital because the incision in her neck was painful and showed outflowing pus. Physical examination showed that the skin around the incision was red, swollen and bulging. The margin of the incision was dehiscent, and the white pus appeared to have a foul odour. Hydrogen peroxide, a metronidazole (MTZ) injection, and saline were used to wash the abscess cavity. A rubber drainage tube was placed through the dehiscence and the wound was irrigated daily with frequent dressing changes. The patient was prescribed oral levofloxacin (200 mg b.i.d.). After one week, the swelling reduced, and the amount of drainage discharge gradually decreased. The drainage tube was replaced with a thinner version and the dressing was changed every three days. In the subsequent days, removal of the drainage tube failed because of the persistent exudates. The patient was readmitted to hospital three months later because of postoperative infection and fistula formation. Fibreoptic bronchoscopy (FOB) showed no obvious abnormalities. Debridement and suturing in the neck wound were performed under general anaesthesia. Intraoperative exploration showed that the sinus tract extended upwards and was in close contact with the left lateral wall of the trachea to the level where the recurrent laryngeal nerve enters the larynx. The surrounding tissue was infected and necrotic. No obvious abscess cavity was observed. The necrotic tissue was removed and surgical exploration confirmed that there was no communication with the tracheobronchial system. The wound was irrigated with hydrogen peroxide and diluted iopromide and a negative-pressure drainage tube was inserted. A week later, the drainage tube was removed. No signs of infection were observed at the patient's six-month follow-up visit.

Case 2

A 75-year-old woman was admitted to hospital complaining of a one-week history of a left-sided cervical painless mass. Physical examination upon admission showed her neck was swollen and a mass (approximately 5 × 4 cm) was palpable in the left thyroid. The mass was painless, pliable, well demarcated, and moveable, without auscultation of vascular murmurs. No palpable nodules or lymph nodes in the right thyroid gland or neck were observed. Ultrasound examination of the thyroid showed a mixed-echoic mass considered to be a goitre with haemorrhage, cystic degeneration, and calcification. Routine blood tests showed an elevated white blood cell (WBC) count

(i.e., 8.9×10^9/l). Results of urinalysis were within normal range as were coagulation, liver, kidney and thyroid function parameters. In addition, electrocardiogram (ECG) thyroid autoantibody and calcitonin levels were normal. A FNAB was not performed due to the critical positioning of the mass.

Two days later, the patient underwent a left thyroid gland lobectomy under general anaesthesia. Intraoperative exploration showed that the left thyroid was adhered to the trachea. A cystic and solid mass measuring approximately 3×3 cm was found in the gland and behind the mass was an irregular bar-like cyst (5×1.5 cm) which was adhered to the trachea. The cyst extended outward to the dorsal end of the left clavicle. Due to its thin wall, the cyst burst on manipulation. To avoid further damage to the oesophagus, trachea, recurrent laryngeal nerve, and apical lung regions, we allowed the wall of the cyst to remain close to the trachea and the left clavicle. We resected most of the wall of the cyst and inserted a drainage tube. Postoperative pathological examination of the tissues showed left thyroid adenoma with cystic change. The cystic mass was lined by columnar epithelium surrounded by layer of smooth muscle with mucus glands. The mass was considered to be a bronchogenic cyst. Three hours after the operation, the patient developed dyspnoea. Anti-asthma therapy and glucocorticoids were given but the patient's general condition worsened. She expectorated brown bloody sputum, and her heart rate was 150 beats/min and respiratory rate was 40 beats/min. Sputum aspiration, a mask for oxygen inhalation, and heart-strengthening medications were given immediately and the patient's condition improved; she was transferred to the intensive care unit (ICU). Wound exploration under general anaesthesia was performed on the following day and a tiny defect on the sidewall of the trachea was found to communicate with the stump of the bar-like cyst. Tracheal repair was performed and symptomatic treatments (i.e., anti-infectives and rehydration therapy) were given postoperatively.

Two days post-surgery, the patient developed facial and thoracic subcutaneous emphysema. Following 48 hours without improvement, wound exploration was undertaken and the tiny defect on the trachea was found to be torn. Considering vocal tissue is fragile, we performed a tracheostomy instead of a defect repair and instigated permanent irrigation and drainage from the wound. Ten days later, the drainage tube was removed and food residues were observed to flow out through the wound. Contrast-enhanced CT showed upper oesophageal fistulas. Following two months of observations with symptomatic treatments, the oesophageal fistulas remained unhealed, so jejunostomy was performed, and enteral nutrition was given. The patient died 125 days after the first operation due to acute upper gastrointestinal bleeding.

Written informed consent for the publication of this report was obtained from the patients or their families and the case reports were authorized for publication by the review board of our institution. This report adheres to CARE guidelines.[9]

Discussion

Reports of ectopic bronchogenic cysts outside the thorax are uncommon and those in the thyroid are exceedingly rare. While some reports describe bronchogenic cysts as asymptomatic, others suggest that the majority of adults with bronchogenic cysts ultimately become symptomatic due to compression of the trachea, oesophagus, and recurrent laryngeal nerve, resulting in dyspnoea, dysphagia, and hoarseness.[4,7,10] In some cases, the cysts may cause secondary infections.[2] Superficial cysts may form sinusoids and drain pus, and deep cysts may

form abscesses.[11] Cyst infections are usually caused by fibrous connections between the cysts and tracheobronchial system. Bacteria can make good use of narrow channels to enter cysts and further cause local or systemic infection and form a deep abscess. Once the channels or abscesses form, they should be surgically removed and the cysts blocked from the tracheobronchial system.[12]

Differential diagnosis of cervical bronchogenic cysts includes other malformations such as, branchial cleft cysts, thyroglossal duct cysts, thymic and thyroid cysts, epidermal and dermoid cysts, cystic hygromas, cystic teratomas, and cystic neuromas.[3] To distinguish these lesions, location and histology are essential. The cysts are characterised by a pseudostratified ciliated columnar epithelium. The cyst wall may also have interspersed areas of smooth muscle, mucus-secreting goblet cells, cartilage, and dystrophic calcification.[3,4] On CT scans, bronchogenic cysts are usually sharply marginated mediastinal cysts, with soft-tissue attenuation, or water attenuation; the majority of them are cystic or cavity-like, and others are solid and thus easily confused.[4,5] When the cysts demonstrate calcification, it is difficult to distinguish them from thyroid adenomas.[13] Magnetic resonance imaging (MRI) with its soft-tissue detail is advantageous for elucidating the cystic nature of these lesions.[14] The lesions show high signals in both T1-weighted and T2-weighted images, probably because of the mixture of water and proteinaceous mucus in cysts. However, while MRI is valuable in demonstrating size, shape and location of cysts, imaging examinations lack specificity in differential diagnoses.[3] With regard to treatment, surgical excision remains the treatment of choice. Cases have been reported in which bronchogenic cysts have caused malignant tumours such as poorly differentiated adenocarcinoma,[15] mucoepidermoid carcinoma,[16] and malignant melanoma.[17] Therefore, surgical excision is recommended since complications are not uncommon.

The two patients reported here had intrathyroidal bronchogenic cysts and developed complications including infection; they subsequently underwent secondary surgical explorations. Given that thyroidectomy is an aseptic operation, the operative region is usually not exposed to the external environment. Therefore, we suspect that connection to the tracheobronchial system was the cause of infection. In hindsight, perhaps the bubbles we noted in the CT images from Case 1 indicated communication between cysts and the tracheobronchial system. If we had noticed this malformation, we probably would have performed careful exploration, repeatedly irrigated the operative field and administered therapeutic antibiotics. For Case 2, perhaps we should have asked our thoracic surgeon colleagues for assistance to reduce the risk of trachea-oesophageal fistulas.

Declaration of conflicting interests

The authors declare that there are no conflicts of interest.

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