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CLINICAL RECORD

Infected tracheocoele (acquired tracheal diverticulum): case report and literature review

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Abstract

Background: Paratracheal air-filled cysts are rare. Tracheocoele or acquired tracheal diverticulum is the term given to these acquired abnormalities, which usually arise in adults. The majority is asymptomatic, being discovered as incidental findings on radiological imaging.

Methods: We report the case of a 72-year-old man with a previously identified tracheocoele which became symptomatic following an upper respiratory tract infection. A literature is presented and nomenclature is discussed.

Results: The clinical presentation, differential diagnosis and management of paratracheal air-filled cysts are discussed.

Conclusion: While most of these rare abnormalities are discovered incidentally, this case illustrates the fact that significant symptoms can develop; excision should therefore be considered.

Key words: Trachea; Diverticulum; Cysts; Diagnosis

Introduction

Air-containing cysts in the paratracheal region were first described in 1838 by Rokitansky as retention cysts of the mucous glands.[1] These cysts are usually found incidentally on chest imaging and are managed conservatively. Their incidence has been reported as 1 per cent on autopsy[2] and 0.75–2 per cent on computed tomography (CT) imaging.[3,4] They have also been reported on fibre-optic bronchoscopy in 0.3 per cent of children aged over 10 years.[5]

As with many rare clinical entities, numerous terms have been used to describe these lesions over the years. These include paratracheal air cyst, tracheal diverticulum, tracheal diverticulosis, intratracheal diverticulum, bronchogenic cyst, aerocoele, bronchocoele and aerial goitre (air-goitre).

By recent convention, the term tracheocoele has become synonymous with acquired tracheal diverticulum.

We present the case of a 72-year-old man with an acquired tracheal diverticulum.

Case report

A 72-year-old Indian man presented with an eight-week history of right lower neck pain and cough productive of yellow sputum. He also complained of night sweats, low-grade fever, weight loss (6 kg in two months) and loss of appetite. His past medical history included chronic obstructive pulmonary disease (COPD), myocardial infarction, a cerebrovascular event and gastroesophageal reflux disease. He had been an ex-smoker for 40 years but had a previous 120 pack-year history.

On examination, there was an expansile mass arising from the right supraclavicular fossa, which increased in size when the patient performed the Valsava manoeuvre. This sign has been previously described in a case of air-goitre.[6]

Five years previously, during a hospital admission for gastroenteritis, a routine chest radiograph and subsequent CT scan had demonstrated an air cyst in the right upper mediastinum ($3.6 \times 2.7 \times 2.5$ cm). The cyst had been seen to cause some displacement of surrounding structures (trachea, right subclavian vessels and common carotid arteries) but otherwise had not displayed any adverse features. The remaining lung fields had been mildly hyper-inflated, with appearances consistent with COPD, but had no other significant abnormalities. Subsequent investigation with flexible nasendoscopy and formal bronchoscopy had failed to reveal any abnormalities. The air cyst had been considered an incidental finding and thus ignored.

During the current presentation, a repeated chest CT scan (Figure 1) demonstrated that the lesion had increased in size, to $4.1 \times 3.4 \times 4.8$ cm. An air–fluid level was now apparent within the cyst. In the lung fields, non-specific pulmonary nodules were seen in the right upper lobe and left lower lobe, together with scarring of the right middle lobe. Other investigations included negative sputum microscopy and microbiological culture (including mycobacterial culture). Arterial blood gases were normal.

The patient's clinical symptoms had initially responded to a short course of antibiotics.

Microlaryngobronchoscopy demonstrated one tract and two further indentations in the posterolateral aspect of the upper trachea at the junction of the trachealis muscle and tracheal cartilages (Figure 2). Pharyngoesophagoscopy was normal.

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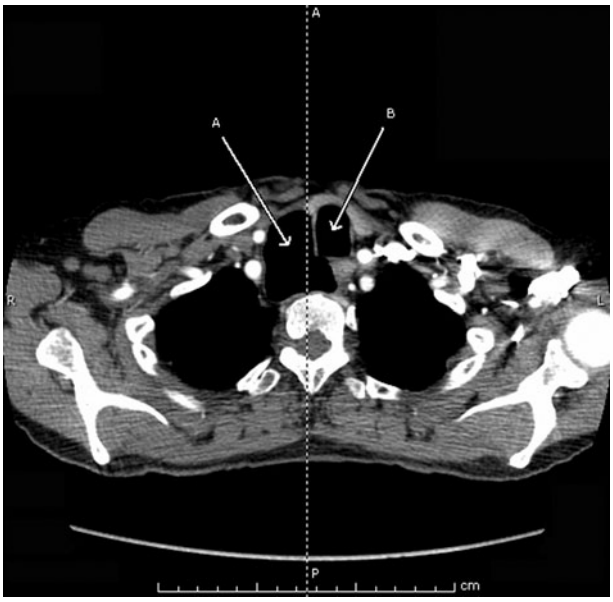

FIG. 1

Typical axial computed tomography appearance of a tracheocoele (A) as a thin-walled cyst in the right paratracheal region (trachea is shown as B). The absence of cartilage within the wall differentiates it from a congenital diverticulum. A = anterior; R = right; L = left; P = posterior

Two days after endoscopy, the patient re-presented with a further significant episode of fever, this time with odynophagia. A repeated CT excluded any post-operative complication. In light of his persistent and recurrent symptoms, a decision was made to undertake surgical resection.

Surgery was performed under general anaesthesia, via a right collar neck incision. The inferior thyroid vessels were ligated and the right lobe of the thyroid mobilised to expose the paratracheal region. The cyst was identified (Figure 3) and a capsular dissection performed. Two fibrous connections to the posterolateral aspect of the trachea were identified and ligated. The recurrent laryngeal nerve was identified, intimately related to the deep surface of the cyst, and was preserved.

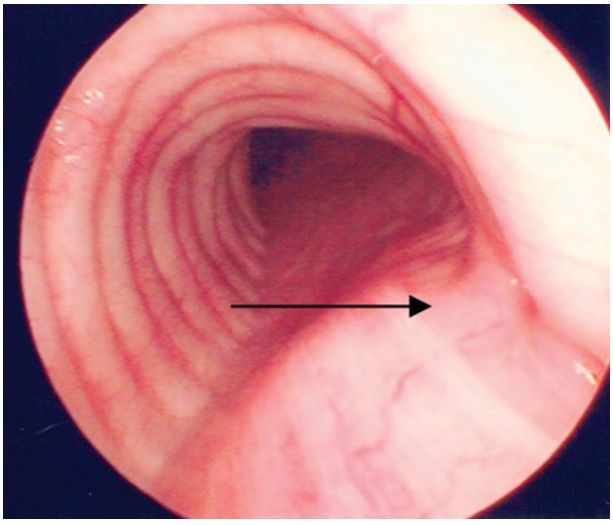

FIG. 2

Endoscopic view showing indentations (black arrow) representing an unusually narrow connection between trachea and tracheocoele.

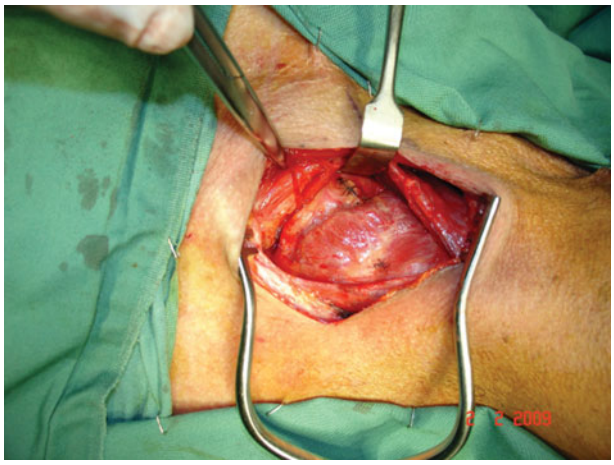

FIG. 3

Operative view showing a thin-walled cyst identified during surgical resection.

The post-operative period was complicated by a temporary ipsilateral recurrent laryngeal nerve palsy, which resolved over nine months.

Histologically, the specimen consisted of a simple cyst lined by ciliated columnar epithelium, with a fibrous tissue wall up to 2 mm thick containing small foci of smooth muscle. The lumen was filled with frothy mucus.

Discussion

We performed a literature review of all English language reports describing paratracheal air-filled abnormalities in adults (Table I). Changing trends in nomenclature made accurate comparison of cases difficult. We therefore separated cases into four groups according to the original description: tracheocoele, tracheal diverticulum, congenital tracheal diverticulum and others (predominantly tracheal diverticulosis). We identified 18 cases described as tracheocoele, 18 as tracheal diverticulum and five as congenital tracheal diverticulum; a further 14 we grouped together as 'other'.

By recent convention,[30,40] those lesions are now defined as tracheal diverticula and sub-divided into congenital or acquired lesions (See Table II).

Acquired tracheal diverticula are true diverticula resulting from mucosal herniation through weak points, most commonly in the right posterolateral trachea (98 per cent) and usually at the level of the thoracic inlet. They are thin-walled and consist of normal respiratory epithelium on a narrow layer of fibrous stroma. These lesions were previously commonly referred to as tracheocoeles.

A congenital tracheal diverticulum represents a malformed, vestigial, supernumerary budding of the trachea.[51] The wall of a congenital diverticulum is similar to the wall of the trachea, containing smooth muscle fibres, cartilage and respiratory epithelium. While congenital tracheal diverticula may occur in isolation, their embryological origins mean they are more likely to occur in association with other upper aerodigestive tract anomalies. These include Mournier–Kuhn disease (congenital dilatation of the trachea and bronchi),[46] tracheoesophageal fistula[52] and tracheobronchomegaly.[40]

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TABLE I
PREVIOUS ENGLISH-LANGUAGE REPORTS OF PARATRACHEAL AIR-FILLED ABNORMALITIES IN ADULTS*

Study	Pt age (y) + sex	Description	Presentation	Surgical approach
Tracheocoele				
Addington *et al.*[7]	32M	Tracheocoele	Choking, coughing, SOB	Nil
Andersen *et al.*[8]	52M	Tracheocoele	Cough productive of large amount of purulent sputum	Sternotomy
Sullivan & Mangiardi[9]	59F	Tracheocoele	Chronic, recurrent, productive cough	Left lateral thoracotomy
Gronner & Trevino[6]	59F	Tracheocoele	Hoarseness	Total laryngectomy & neck dissection (for concurrent laryngeal SCC)
Scholl[10]	79F	Tracheocoele	Globus sensation	Nil
Moller *et al.*[11]	90F	Tracheocoele	Difficult intubation	Nil
Henderson *et al.*[12]	50F	Tracheocoele	During tracheostomy for trauma	Oversewing of communication with trachea
Mathur *et al.*[13]	50M	Tracheocoele	Progressive right neck swelling	Y-shaped neck incision
Grassi *et al.*[14]	44M	Tracheocoele	Pain in anterior neck	Nil
	74M	Tracheocoele	Coughing paroxysms & recurrent chills	Nil
	86M	Tracheocoele	Acute respiratory infection	Nil
Piazza *et al.*[15]	27M	Tracheocoele	Left paratracheal swelling noticed during assisted ventilation	Nil
Endo *et al.*[16]	78F	Tracheocoele	Cough, neck irritability; incidental finding on pre-operative CT for hemithyroidectomy	Neck collar incision prior to hemithyroidectomy
Porubsky & Gourin[17]	58M	Tracheocoele	Intermittent productive cough, dysphagia, worsening dyspnoea	Transverse cervical approach
Teker *et al.*[18]	67M	Tracheocoele	Pain & swelling to right of clavicle	Resection of cyst then repair of posterior tracheal wall
Shah *et al.*[19]	50F	Tracheocoele	Neck swelling, pulling sensation, intermittent hoarseness & SOB	Nil
Danielson *et al.*[20]	81M	Tracheocoele	Chronic cough following retropharyngeal abscess	Nil
Yazkan *et al.*[21]	66M	Tracheocoele	Intermittent chest pain aggravated by exercise	Nil
Tracheal diverticulum				
Nielsen[22]	51M	Tracheal diverticulum	Recurrent lung infections, copious expectoration decreased after postural drainage	Nil
Surprenant & O'Loughlin[23]	28M	Tracheal diverticulum	Right frontal headache, rhinitis, productive cough, fever & chest pain (tracheobronchomegaly)	Nil
Infante *et al.*[24]	59M	Tracheal diverticulum	Chronic productive cough, coughing fits, stridor	Right lateral cervicotomy
Caversaccio *et al.*[25]	70F	Tracheal diverticulum	Severe dyspnoea, stridor, dysphagia, recurrent laryngeal nerve paralysis	Nil
Davies[26]	77F	Tracheal diverticulum	Difficult tracheal intubation	Nil
Ching *et al.*[27]	67M	Tracheal diverticulum	Difficult lung isolation during intubation	Nil
Rahalkar *et al.*[28]	47F	Tracheal diverticulum	Chronic cough, dysphonia	Nil
Narimatsu *et al.*[29]	59M	Tracheal diverticulum	Incidental finding on CT post-trauma	Nil
Soto-Hurtado *et al.*[3]	49M	Tracheal diverticulum	Incidental finding on CXR	Nil
	63F	Tracheal diverticulum	Haemoptysis	Nil
Ampollini *et al.*[31]	77M	Tracheal diverticulum	Incidental findings on CT	Nil
Modrykamien *et al.*[32]	45F	Tracheal diverticula	Fever, productive cough, constitutional symptoms (history of cystic fibrosis)	Bilateral lung transplant, tracheal diverticulum managed conservatively
Han *et al.*[33]	50M	Tracheal diverticulum	Mild dysphagia, sensation of friction	Right semi-collar incision
Kokkonouzis *et al.*[34]	62M	Tracheal diverticulum	Chronic cough	Nil
Morgan *et al.*[35]	40M	Tracheal diverticulum (paratracheal air collection)	Incidental finding on CXR & CT post-trauma	Nil
Pinot *et al.*[36]	34M	Acquired tracheal diverticulum	Productive cough, haemoptysis	Nil
Haghi *et al.*[37]	72M	Tracheal diverticulum (paratracheal air cyst)	Productive cough, haemoptysis, exertional dyspnoea	Nil
Sato *et al.*[38]	65F	Tracheal diverticulum (paratracheal air cyst)	Throat & neck pain	Nil
Congenital tracheal diverticulum				
Peytz[39]	50M	Congenital tracheal diverticulum	Severe cough	Nil
Sharma[40]	50F	Tracheal diverticulum	Incidental finding on CXR	Nil
	55F	Tracheal diverticulum	Incidental finding on CXR	Nil
	35F	Tracheal diverticulum	Incidental finding on CXR post-trauma	Nil

Continued

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Table I *Continued*

Study	Pt age (y) + sex	Description	Presentation	Surgical approach
Oizumi *et al.*[41]	66M	Congenital bronchial diverticulum	Positional localised wheezing	Nil
Other				
Morlock & Pinchin[42]	50M	Bronchial diverticulosis	Productive cough	Nil
Goldman & Wilson[43]	43M	Tracheal diverticulosis	'Chicken bone in throat', chronic non-productive cough	Foreign body removed, tracheal diverticulosis managed conservatively
Ettman & Keel[44]	40M	Tracheal diverticulosis	Acute respiratory infection	Nil
	32M	Tracheal diverticulosis	Productive cough, fever, SOB	Nil
	43M	Tracheal diverticulosis	Productive cough, SOB on exertion	Nil
Collins & Wight[45]	67F	Posterior tracheal wall diverticula (both pts)	Incidental finding during total laryngectomy (both pts)	Nil
	50M			Nil
Lazzarini de Oliveira *et al.*[46]	38M	Tracheal diverticulosis	Chronic productive cough (Tracheomegaly, bronchiectasis)	Nil
Kim *et al.*[47]	43F	Paratracheal air cyst	Incidental finding on neck US	Nil
	65M	Paratracheal air cyst	Anterior neck pain	Nil
Saito *et al.*[48]	74M	Tracheobronchial diverticula	Productive cough, right middle lobe atelectasis	Nil
Levin *et al.*[49]	19M	Tracheal diverticulosis	Worsening SOB & productive cough in congenital HIV pt	Nil
Hernández Pérez *et al.*[50]	44M	Intratracheal diverticulum	Dyspnoea, cough, haemoptysis, stridor	Nil
Sharma[40]	20M	Tracheal diverticulosis	Cough, dyspnoea, fever (congenital tracheobronchomegaly)	Nil

*Older than 18 years. Pt = patient; y = years; M = male; F = female; SOB = shortness of breath; SCC = squamous cell carcinoma; CT = computed tomography; CXR = chest X-ray; US = ultrasound; HIV = human immunodeficiency virus

TABLE II
DISTINGUISHING FEATURES OF CONGENITAL AND ACQUIRED TRACHEAL DIVERTICULA

Parameter	Congenital tracheal diverticulum	Acquired tracheal diverticulum*
Aetiology	Congenital developmental defect	Acquired mucosal herniation usually associated with raised intra-luminal pressure
Size	Small	Large
Site	Posterior (4–5 cm below carina)	Right posterolateral (thoracic inlet)
Structure	Multiple sacs Narrow communication	Single sac Wide communication
Histology	Complete tracheal anatomy (respiratory epithelium, smooth muscle & cartilage)	Thin-walled, fibrous cyst lined with normal respiratory epithelium
Contents	Mucus	Predominantly air-filled
Associated conditions	Tracheoesophageal fistula	Chronic obstructive airway disease

*Also known as tracheocoele

Tracheal diverticula have also been reported in association with: trachiectasis (dilatation of the trachea with multiple small herniations of the membranous portion of the trachea),[8] congenital human immunodeficiency virus infection,[49] cystic fibrosis[32] and Duchenne muscular dystrophy.[15]

Other air-filled abnormalities can occur at the thoracic inlet. The differential diagnosis includes lymphoepithelial cyst, bronchogenic cyst, laryngocoele, pharyngocoele, Zenker's diverticulum, pneumomediastinum, apical hernia of the lung, and apical paraseptal blebs or bullae.[4,18,30]

Acquired tracheal diverticula are thought to occur as a result of a persistently raised intratracheal pressure associated with chronic respiratory conditions. They occur in pre-existing areas of potential weakness in the posterolateral tracheal wall between the tracheal cartilage and trachealis muscle. They occur almost exclusively on the right side, probably due to the relative positions of the trachea and oesophagus on the left. Previous reports suggest that they should have relatively wide connections to the trachea, compared with congenital diverticula; however, this was not the case in our patient.

It is likely that the majority of cases are asymptomatic, being discovered incidentally on routine radiological investigation, including X-ray,[30,40] CT[16,31] and ultrasound.[47] Most reported cases are symptomatic, usually due to a local mass effect and direct compression, resulting in cough, dyspnoea, stridor, dysphagia, and chest, neck and/or right clavicular pain. Symptoms can also arise due to vagal irritation (resulting in cough, dysphonia or vocal fold paralysis) or alternatively due to retained secretions (serving as a reservoir of chronic infection) or recurrent irritation of the upper airways. Tracheal diverticula have also been found incidentally during tracheostomy,[12] laryngectomy,[45] difficult intubation,[11,26,27] and following trauma.[40,29,35]

CT scanning has been shown to be the most effective method for evaluating the presence and features of tracheal diverticula.[53] In most cases, a CT scan and barium swallow will exclude the differential diagnoses listed above.

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Previous authors have suggested that surgery is only indicated in young, symptomatic patients in whom conservative measures (e.g. antibiotics, mucolytics and physiotherapy) have been unsuccessful. The current case demonstrates that asymptomatic lesions can become problematic over time. It also demonstrates that age and other pre-existing co-morbidities should not be considered as absolute contraindications to surgery.

- **Tracheal diverticulum is a rare anomaly of the tracheobronchial tree**
- **The nomenclature describing paratracheal air cysts is confusing**
- **Such lesions are now divided into congenital and acquired tracheal diverticula (the latter also known as tracheocoeles); these subtypes have different aetiologies and characteristics**
- **This case report illustrates the necessity of surgical resection in cases of symptomatic tracheal diverticulum**

Where surgery is considered, the most commonly described approach is resection via a transverse or lateral neck incision.[17,24] Other reported options have included fulguration,[54] endoscopic cauterisation with laser or electrocoagulation,[30,32] and endoscopic division with biopsy forceps.[55]

Conclusion

Most of the cases identified in our literature review, other than those reported as congenital tracheal diverticula, probably represent similar clinical entities. However, an inconsistent approach to the naming of these lesions makes accurate comparison of previously reported cases difficult. With regards to the clinical management, it is important to identify congenital lesions so that associated abnormalities can be investigated and managed appropriately. For the remaining acquired lesions, any further sub-classification has little bearing on clinical decision-making. We would therefore advocate the simple classification of cases into congenital or acquired lesions. For acquired lesions, surgery should be considered in symptomatic cases in which simple medical treatment options have been unsuccessful.

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