Tracheocele Presenting with Intermittent Dysphonia: A Case Report

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Received: 9 November 2011 / Accepted: 9 February 2012 / Published online: 4 March 2012
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Abstract We report a rare case of tracheocele presenting in an ENT setting. The referral was made on the basis of intermittent dysphonia. The aim of this report is to document the rare condition of tracheocele on the right side and to help raise the level of its awareness among the otolaryngologists. So far approximately thirty cases of this condition have been documented in the literature worldwide. An emphasis is placed on the mode of presentation and the management issues, as early diagnosis is crucial and offers a favorable prognosis. The right sided predilection of the swelling is due to anatomical reason and the cause of recurrent dysphonia is explained.

Keywords Neck swelling · Lymphangioma · Branchial cyst · Veinectasis · Pulmonary bulla · Phonasthenia

Case Report

A previously well 48-year-old gentleman working in a telecom sector presented with a complaint of a swelling on the right side of the neck above the sterno-clavicular joint. Which occurred abruptly after a loud telephonic conversation 2 months ago, subsequently patient developed change in voice whenever the swelling increased in size

during conversation on telephone. There was no prior history of change in voice, cough, dysphagia or trauma to the neck. The right side neck swelling (Fig. 1) was about the size of a ping pong ball which increased in size on straining and reduced on compression, accompanied by audible hissing noise. Further examination with flexible fiberoptic endoscope, showed a normal structure and movements of vocal cords. Further tracheoscopy revealed a mild bulge of the right lateral tracheal wall, and distal to the bulge, the rest of the trachea up to carina was unremarkable. On manual compression of the neck swelling, air bubbles were seen over the right postero-lateral wall of the trachea below the cricoid ring but no definite opening could be seen in the trachea. High resolution CT scan of the neck and chest taken, (Fig. 2) revealed an air filled, loculated sac measuring around $5 \times 4 \times 3$ cms with a small communication at the level of the T1 vertebra. Anterior-posterior and lateral view of the reconstructed pictures showed the communicating tract clearly, confirming the diagnosis of a tracheocele (Fig. 3). Patient was advised surgical excision.

Surgery

The swelling was approached through a transverse collar incision. By subplatysmal dissection, the sternocleidomastoid muscle was retracted laterally and strap muscles medially; the sac was identified which looked like a ranula (Fig. 2). The sac was made to bulge with increasing in tracheal pressure by mechanical ventilation by the anaesthetist. The sac while being dissected meticulously, the right recurrent laryngeal nerve was seen coursing over the sac. The nerve was separated and retracted medially, avoiding injury (Fig. 4). The neck of the sac was identified tracking down to the third and fourth tracheal rings. The

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Fig. 1 Right neck swelling

Fig. 2 Sac exposed-looking like a ranula

Fig. 3 Right recurrent laryngeal nerve coursing over the sac

Fig. 4 HPE: cyst lined by pseudo stratified columnar epithelium (H&E $\times 200$)

sac was opened and found to be multilocular without any contents inside. The sac was dissected up to the tracheal wall on the right side. The neck of the sac was found

Fig. 5 HRCT neck-sac with communication

Fig. 6 HRCT-reconstructed image-P.A view

entering the trachea, and after pulling the sac outwards by a clamp, titanium Ligaclip was applied close to neck, near the tracheal wall (Fig. 5). Then sac was excised and sent for histopathological examination. Water test was done to check for any leak. After obtaining complete hemostasis, the wound was closed in layers without a drain. Patient had uneventful recovery and skin clips were removed on the seventh post-operative day. Histopathology report confirmed tracheocele (Fig. 6).

Discussion

Tracheocele is an air cyst due to the protrusion of the lining membrane of the trachea, due to structural weakness. Acquired tracheoceles are rare, as fewer than thirty cases have been reported in the world literature [1–4]. In adults tracheoceles are acquired, according to some the cause may be due to mucus gland obstruction or due to ectatic changes in the tracheal wall, which is the area of weakness and increase in intra-luminal pressure results in tracheoceles [5, 6]. There is a weak area between the transverse bands of the trachealis muscle through which the mucosa may herniate secondary to increased intra-luminal pressure [7].

Dilatation of the trachea due to multiple herniations of the membranous portion is called trachiectasis, also called tracheal or bronchial diverticulosis [8–12] or multiple diverticular tracheomalacia [13]. On rare occasions, this condition has been associated with the formation of tracheoceles which has been referred to as tracheal diverticulum, bronchocele, arocele or air-cyst [1]. A tracheocele may be present without associated trachiectasis [1]. In this patient reported there is no evidence of trachiectasis on bronchoscopy.

The etiology of this condition is unknown, but in the instances reported the development of the symptoms during adulthood leads one to believe they are acquired and not congenital tracheoceles. The pathological feature is destruction of elastic tissue as in bronchiectasis. The histopathology report is only an air-cyst with respiratory epithelial lining (Fig. 4).

My patient reported on 5th February 2011 for a review. He had no recurrence of the swelling and voice fatigue on prolonged conversation has disappeared but the quality of voice remains the same. On fibre-optic flexible laryngoscopy, revealed the right vocal cord was in paramedian position with well compensated left vocal cord movement and there is no phonatory gap. He is advised voice therapy and periodic review.

Most of the tracheoceles have a predilection to the right side of the trachea, probably as a result of lack of the positional support of the wall of the esophagus [7]. There is only one such case of tracheocele on the left side, reported till date [14].

The involvement of the recurrent laryngeal nerve has been reported due to the expansion of the swelling causing total dysphonia, but there are no case reports of intermittent dysphonia which happened in our patient due to intermittent pressure on the nerve by the expanding swelling. The radiological findings of the tracheocele have been described in several reports but few operative findings have been documented [4, 13, 15–23]. These swellings have to be differentiated from lymphangioma, branchial cyst, venectasis and pulmonary bulla by clinical examination and

radiological investigations [24]. Surgery is mandatory if it is symptomatic.

Conclusion

This case highlights the importance of clinical findings corroborating with endoscopic and radiological features associated with right sided compressible swelling. The accepted treatment is excision of the swelling if symptomatic. In this case surgery was imperative because the patient was conscious of neck swelling and voice fatigued on prolonged conversation which was due to building up of air pressure, increasing the size of the swelling and exerting pressure on the recurrent laryngeal nerve. The clinical diagnosis supported by the radiological findings helped in the surgical approach. Untreated tracheoceles may cause recurrent infection, aspiration pneumonia, mass effect, dysphagia, vocal fold paralysis, and pneumomediastinum.

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