

Laryngeal cyst of unusual origin and presentation

M Bochnia, M Fr?czek, J Rabczy?ski

Citation

M Bochnia, M Fr?czek, J Rabczy?ski. *Laryngeal cyst of unusual origin and presentation*. The Internet Journal of Otorhinolaryngology. 2007 Volume 8 Number 1.

Abstract

Branchial cysts are uncommon developmental anomalies rarely affecting the larynx. They arise mostly from the second branchial pouch, largely in the left anterior side of the neck. The peak incidence rate occurs in childhood. The most frequent clinical findings are voice disorders and respiratory distress of varying intensity. We describe a case of unusual presentation of IV branchial anomaly imitating a laryngeal cyst of typical epithelial origin. The lesion is reported also for its right-side location, advanced patient's age and long, uneventful history. The authors point out the need to consider also such a rare anomaly among other more frequent intralaryngeal lesions.

INTRODUCTION

Laryngeal cysts are rare, mostly benign lesions affecting both sexes and all age groups. The symptoms depend on cyst localization and size. Smaller laryngeal cysts located inside the epilarynx are mostly asymptomatic. The commonly presenting symptoms of larger lesions are voice disorders (hoarseness and dysphonia), dysphagia and even airway obstruction with inspiratory stridor. Dyspnoe is extremely dangerous among new born infants and can even result in tracheotomy in severe cases.

Laryngeal cysts categorization is based mostly on assumptions on their origin and localization [1]. Most of them distinguish the group of so called epithelial cysts including saccular and ductal. Lateral and anterior saccular cysts originate from the saccula, being the tiny appendage of the laryngeal ventricle. These lesions are the counterparts of the adult laryngoceles. Ductal cysts result from retention of mucus in dilatated collecting ducts of the submucosal glands. They can arise anywhere that a mucous producing gland exists. Among malformations in the larynx, being frequently not included as a separate entity, belong branchial cleft cysts [2].

We would like to describe herein one of the most uncommon of them. This is an example of fourth branchial cleft cyst of untypical localization and clinical presentation.

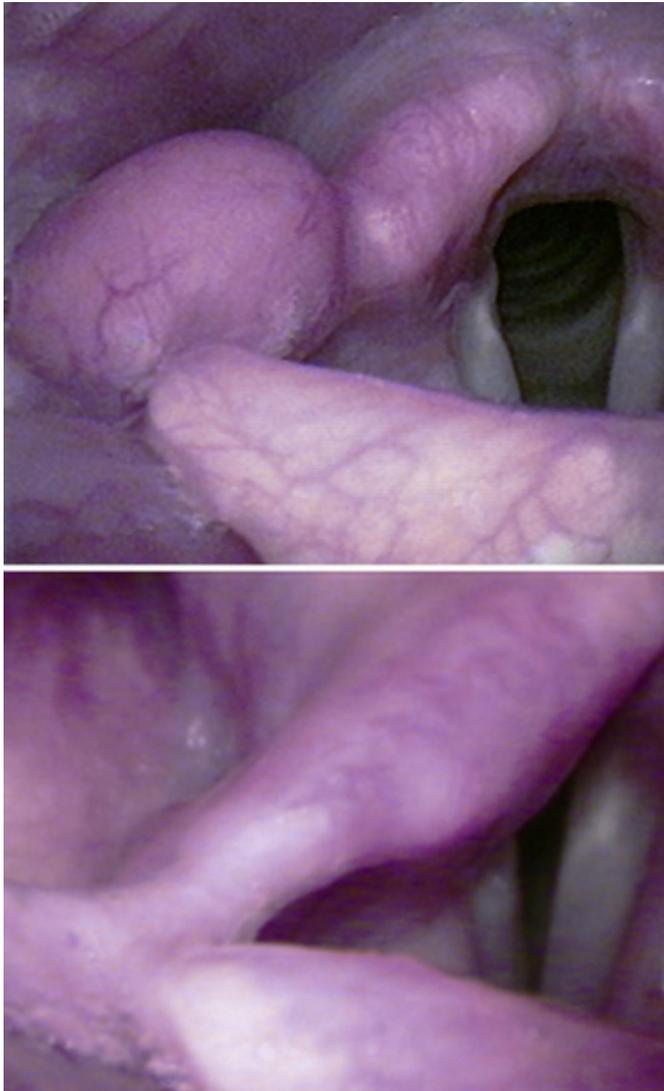
CASE REPORT

An 55-year-old woman working for 26 years as a librarian was originally presented to a private otolaryngologist with a

six-month history of slight hindrance in the pharynx. Indirect laryngoscopy revealed a smooth-surface oval mass about 1 cm diameter located on the right arytheno-epiglottic fold (Fig 1).

Figure 1

Figure 1: Endoscopic view of a branchial cyst located on the right arytheno-epiglottic fold before (upper panel) and one year after surgical excision (lower panel)

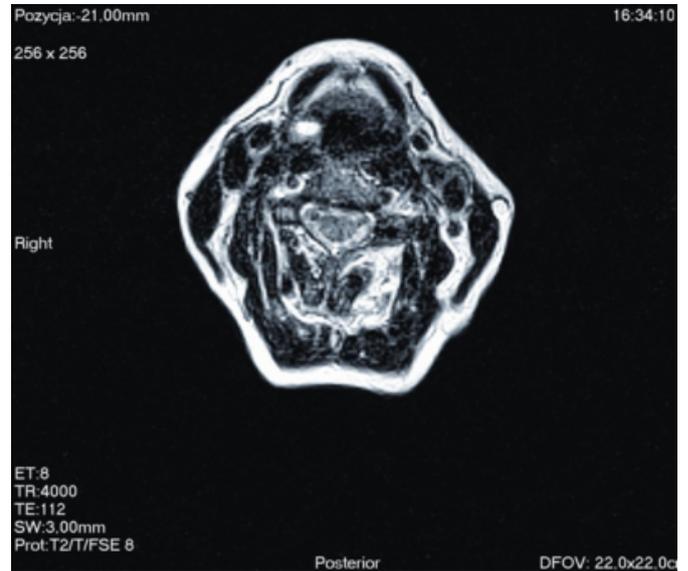


The rest of the examination was unremarkable. The patient was then admitted to the Department of Otolaryngology, Wroclaw Medical University for further evaluation and treatment.

Magnetic Resonance Imaging (MRI) of the head and neck was performed and revealed a unilocular mass showing enhanced signal intensity with no other abnormalities (Fig 2).

Figure 2

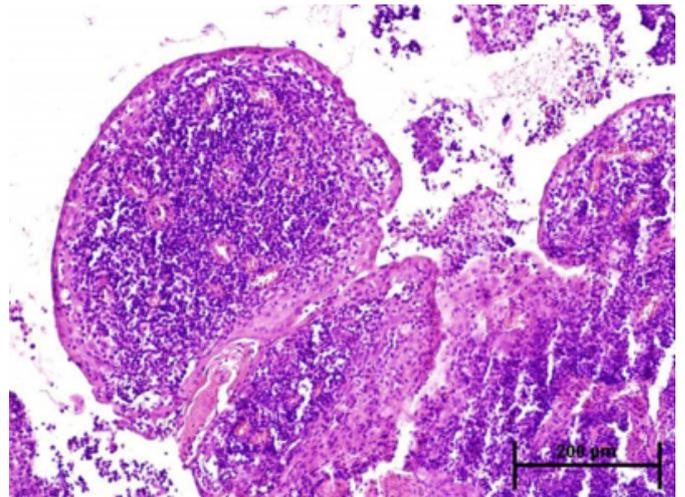
Figure 2: T2 weighted axial image shows high signal intensity anomaly in the region of left arytheno-epiglottic fold



The patient was qualified for surgical intervention. Using suspension laryngoscopy with endolaryngeal approach the cyst was excised entirely. Histopathologic examination of the surgical specimen proved the removed mass was a branchial cleft cyst (Fig. 3).

Figure 3

Figure 3: Cross section of the cyst wall typically lined with stratified squamous epithelium and rich lymphoid infiltrate arranged in a follicular pattern beneath (H&E stain; original magnification x140)



The postoperative course was uneventful and the patient was discharged from the hospital on the second day. The follow-up visit one year after therapy revealed no local recurrence so far.

DISCUSSION

According to the most widely accepted theory branchial cysts, sinuses and fistulas are the results of various degrees of incomplete involution of the branchial apparatus. In utero, both 3rd and 4th pouches originate in the primitive pharynx, from what will become the pyriform fossa. Failure of the surrounding mesenchyme to obliterate their lumen leads to a persistence of these anomalies [3].

The fourth arch remnants are rarest among branchial malformation, and only a few reports are available in the literature [4,5] with the first reported case by Tucker and Skolnick [6]. The location of the malformation is critical in distinguishing 3rd from 4th pouch anomaly. One of the criteria that have been used to differentiate 3rd from 4th pouch sinus or fistula is the position of their internal opening in the pyriform fosse (3rd cephalad; 4th at apex, ie caudal part of the fosse, or even in the proximal esophagus) [7]. Third pouch remnant's ducts are described also as passing superior to the superior laryngeal nerve and posterior to the common carotid artery. Fourth pouch remnants should emerge caudal to the thyroid cartilage and cricothyroid muscle and pass between the superior and recurrent laryngeal nerves [8]. Lateralization of both pouch lesions was found to the left side of the body probably because of embryological asymmetry. The rare presence of right side anomaly is difficult to explain. In the literature cases of bilateral remnants [9] and a small number of right-sided changes have been reported [8, 10,11]. The location of the presented above cyst neighbored with common sites of 4th pouch right side anomaly orifice. According to the authors' knowledge it is a first report of an example of this congenital malformation located in the larynx so superficially on the right side.

Male and females seems to be equally affected by laryngeal cysts [12]. According to the literature, these lesions have peak incidence rate in the third decade of life, appear in the upper one-third of the neck and have a hereditary tendency [12]. There are only a few documented cases of branchial cysts diagnosed later than after the fourth decade [13,14,15]. It is proposed that with time some branchial pouch anomalies can disappear after several episodes of infection by the destruction of the embryonic tissue and the lumen obliteration. This could be a reason for the limited size of

mentioned cysts. The small number of upper airway infections in the presented patient may explain the preservation of the cyst until maturity. Due to the localization and small size described, the malformation was almost asymptomatic for five decades.

In conclusion we would like to point out that the small oval arytheno-epiglottic fold cyst appeared to be a very rare congenital malformation. The presented case emphasizes also the need to consider lesions of 4th branchial pouch origin despite its superficial, right side location, asymptomatic course and advanced patient's age.

References

1. Newman DH, Taxy JB, Laker HI. Laryngeal cysts in adults: a clinicopathologic study of 20 cases. *Am J Clin Pathol* 1984;81:715-20.
2. DeSanto LW. Laryngocele, laryngeal mucocele, large saccules, and laryngeal saccular cysts: a development spectrum. *Laryngoscope* 1974;84:1291-96.
3. Chandler JR, Mitchell B. Branchial cleft cysts, sinuses, and fistulas. *Otol Clinics North Am* 1981;14:175-186.
4. Sharma HS, Razif A, Hamzah M, Dharap AS, Mahbar Z, Kamal MZM. Fourth branchial pouch cyst: an unusual cause of neonatal stridor. *Int J Pediatr Otorhinolaryngol* 1996;38:155-161.
5. Shrimme M, Kacker A, Bent J, Ward RF. Fourth branchial complex anomalies: a case series. *Int J Pediatr Otorhinolaryngol* 2003;67:1227-1233.
6. Tucker HM, Skolnick ML. Fourth branchial cleft (pharyngeal pouch) remnant. *Trans Pa Am Acad Ophthalmol Otolaryngol* 1973;77:368-371.
7. Liberman M, Kay S, Emil S, Flageole H, Nguyen LT, Tewfik TL, Oudjhane K, Laberge JM. Ten years of experience with third and fourth branchial remnants. *J Pediatr Surg* 2002;37(5):685-690.
8. Rosenfeld RM, Biller HF. Fourth branchial pouch sinus: diagnosis and treatment. *Otolaryngol Head Neck Surg* 1991;105:44-50.
9. Rossiter JL, Topf P. Acute suppurative thyroiditis with bilateral pyriform sinus fistulae. *Otolaryngol Head Neck Surg* 1991;105:625-627.
10. Lin JN, Wang KL. Persistent third branchial apparatus. *J Pediatr Surg* 1991;26:663-665.
11. Makino S, Tsuchida Y, Yoshioka H. The endoscopic and surgical management of pyriform sinus fistulae in infants and children. *J Pediatr Surg* 1986;21:398-401.
12. Mandell DL. Head and neck anomalies related to the branchial apparatus. *Otolaryngol Clin North Am* 2000;33(6):1309-1332.
13. Telander RL, Deane SA. Thyroglossal and branchial cleft cysts and sinuses. *Surg Clin N Am* 1977;57:779-91.
14. Ramirez-Camacho R, Garcia Berrocal JR, Borrego P. Radiology Quiz Case 2. *Arch Otolaryngol Head Neck Surg* 2001;127:1395-6.
15. Hart C, Opperman DA, Gulbahce E, Adams G. Branchial cleft cyst: A rare diagnosis in a 91-year-old patient. *Otolaryngol Head Neck Surg* 2006;135:955-957.

Author Information

Marek Bochnia

Professor, Department of Otolaryngology, Faculty of Dentistry, Wrocław Medical University

Marcin Fr?czek, MD, PhD

Department of Otolaryngology, Wrocław Medical University

Jerzy Rabczy?ski

Professor, Department of Pathological Anatomy, Medical University