

## A branchial cyst of the pyriform fossa transoral laser resection: a case report

Hesham Mostafa Abdelfattah · Mohammed Elrabie Ahmed ·  
Mona El-Rabie Ahmed · Mohamed Abd El-Kader Ahmed ·  
Abd-Elmateen Moussa

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**Abstract** Pyriform sinus malformations represent rare third and fourth branchial anomalies. Fistulae at the latter site were initially described and make up less than 1 % of all brachial anomalies. They may be discovered incidentally, or may present as a neck mass with recurrent infection, dysphagia, or airway compromise, and can be an unusual cause of dysphonia in infant and children. Here, we present a case of third branchial cyst located in pharyngeal wall of the left pyriform sinus which presented with dysphonia since birth in a 6-year-old girl. Transoral CO<sub>2</sub> laser excision was carried out successfully with no communicating tract. The patient's dysphonia showed progressive regression at 1-year follow-up. Third branchial cyst in the left pyriform sinus (Bailey's type IV) is an unusual cause of dysphonia in pediatric. Our present case report is the first brachial cyst to be reported in the

pyriform fossa and the second branchial anomalies to be excised transorally with CO<sub>2</sub> laser.

**Keywords** Branchial · Cyst · Pyriform fossa · Transoral · Laser

### Introduction

Pyriform sinus fistula (PSF) and malformations represent rare third and fourth branchial anomalies. They are an unusual cause of dysphonia in infant and children. Pyriform sinus fistulae were initially described and making up less than 1 % of all brachial anomalies [1, 2]. The sinus tract originates in the pyriform (or piriform) fossa. Hence, these anomalies have been called pyriform sinus "fistulae" even though an external opening to the skin rarely is present. These remnants almost always occur on the left side, although a small number of right-sided anomalies have been reported [2, 3].

The branchial cysts (BCs) may be associated with a sinus or a fistula. A cyst with extending sinus is the most common. About 95 % arise from the second branchial arch, while 5 % arising from the first, third or fourth arches, combined [4].

BCs have been divided into four subtypes according to Bailey's classification [5]. Type I cysts are superficial along the anterior border of the sternocleidomastoid muscle beneath the cervical fascia. Type II cysts lie on the great vessels, while type III pass between the great neck vessels to the lateral pharyngeal wall. Type IV is a columnar-lined cyst that lies in the pharyngeal mucosal space and medial to the great neck vessels. Most branchial cysts consist of types 1, 2 and 3; whereas type 4 cysts are extremely rare [6–10].

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H. M. Abdelfattah  
Otorhinolaryngology-Head and Neck Surgery Department,  
University of Alexandria, Alexandria, Egypt  
e-mail: hisham\_fattah@yahoo.com

M. E. Ahmed (✉) · M. A. Ahmed · A.-E. Moussa  
Otorhinolaryngology-Head and Neck Surgery Department,  
Faculty of Medicine, Sohag University, Nasr City, Eastern  
Avenue, University Street, Sohag, Egypt  
e-mail: dr.mohammedelrabie@gmail.com

M. A. Ahmed  
e-mail: Mabelkader1964@gmail.com

A.-E. Moussa  
e-mail: abdelmateen2004@yahoo.com

M. El-Rabie Ahmed  
Phoniatrics, Department of Otorhinolaryngology-Head and Neck  
Surgery, Sohag University, Sohag, Egypt  
e-mail: daimahmoud@yahoo.com

Many theories have been proposed to explain the origin of BCs. The “branchial apparatus remnant theory” which is the most accepted [11, 12]. It suggests that the lining epithelium of the branchial cyst is derived from branchial cleft ectoderm or branchial arch/pouch endoderm (pseudostratified columnar epithelium) or both epithelial types as in case of a long-standing cyst.

On other hand, the “cervical sinus theory,” postulates that BCs represent the remains of the cervical sinus which is formed when the second branchial arch grows caudally to meet the fifth [13, 14]. The third theory is the “thymopharyngeal duct theory” which suggests that these cysts are the remnants of the original connection between the thymus and the third branchial pouch from which it arises [15, 16]. A fourth one is the “lymph node inclusion theory” suggests that these cysts are a result of cystic alteration of epithelium trapped in the cervical lymph nodes [13, 17].

We now report a case of third branchial cyst located in pharyngeal wall of the left pyriform sinus.

### Case report

A 6 year-old girl presented with dysphonia since birth which is stationary in course and not associated with other complaints as breathing and/or swallows. Clinical examination revealed that there is anterior neck swelling measuring  $2 \times 2$  cm at maximum diameter (Fig. 1).

Flexible laryngoscopy revealed left vocal fold immobility near midline position with normal right vocal fold mobility. Also, a cystic swelling arising from medial wall



**Fig. 1** Midline neck swelling, subcutaneous epidermoid cyst (circle)

of left pyriform fossa was visible. It measured about  $3 \times 2$  cm with the arytenoids tilting towards its maximum diameter (Fig. 2). Computed tomographic imaging revealed midline subcutaneous cystic swelling and submucosal cyst in left pyriform fossa (Fig. 3).

Patient underwent transoral CO<sub>2</sub> laser excision under general anesthesia. A submucosal cystic mass located in medial wall of left pyriform sinus where dissection from pharyngeal musculature was done. The mass was excised completely with no communicating tract. Nasogastric tube (NGT) placement was used for feeding after surgery for 7 days. Broad spectrum antibiotics were described for 2 weeks and proton pump inhibitors for 2 months (Fig. 4). Histopathologic evaluation revealed a branchial cyst lined by pseudostratified columnar ciliated epithelium (Fig. 5).

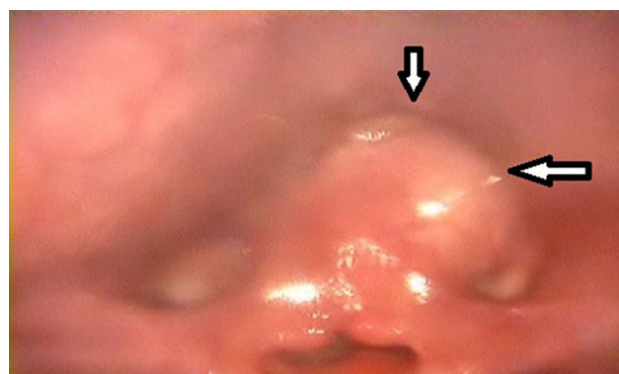
Three months later, the patient underwent surgical excision of the midline cystic neck swelling. It was subcutaneous not attached to surrounding and accidentally opened revealed keratin contents. Histopathologic evaluation revealed epidermoid cyst.

She was scheduled for regular follow-up every month in the first 3 months and every 3 months for 1 year. The patient showed post operative progressive regression of dysphonia.

### Discussion

In 1855 (20 years after Rathke’s startling discovery of the existence of the branchial clefts in mammalian embryos), Langenbeck described two cases of neck cysts which diagnosed as persistent branchial remnants. Three years later, Virchow described another case [5].

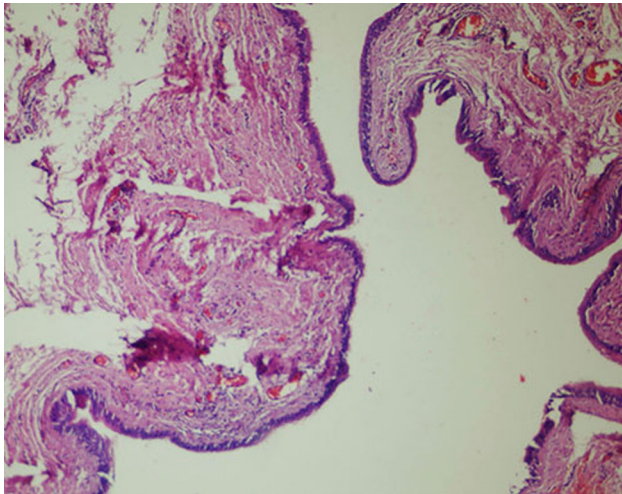
Third and fourth branchial anomalies may be presented at any age [18, 19]. Recently, Ahn et al. [20] reported 21 cases of pyriform sinus fistulae, with pediatrics to adults ratio = 5:4, with the ratio of third: fourth branchial anomaly = 3:9 in pediatrics and 2:7 in adults.



**Fig. 2** Left pyriform cyst sitting piggyback on the left arytenoids (arrows)



**Fig. 3** CT scan finding with well-defined cyst (*arrows*)



**Fig. 4** Histopathological findings of cyst excised



**Fig. 5** Follow-up 2 months after transoral laser excision

It may be more dangerous in neonates because of rapid enlargement causing tracheal compression and respiratory distress. The common presentations include history of recurrent upper respiratory tract infection, neck or thyroid pain and tenderness, and neck mass. Other presentations include cellulitis, thyroiditis, abscess, odynophagia, hoarseness and stridor [21]. Also, it may present as cold thyroid nodules in noncommunicating or noninfected communicating cysts [22, 23], since in utero the pharyngobranchial duct lies in or nearby the thyroid gland. A combination of ultrasound and CT with or without oral contrast will assist in the diagnosis [24].

The present case is the first case report in literature to describe a branchial cyst in the pyriform fossa; although it has been suggested by Bailey in 1923 [5]. Branchial fistulae in the latter location have abundantly reported in the literature.

Laser excision of third and fourth branchial arch remnants was reported only once for 3rd pharyngeal pouch [25]. So, this is the second case that excised by transoral CO<sub>2</sub> laser and the first (Bailey's type IV) third branchial cyst reported in the literature.

Endoscopic technique has evolved as the mainstay of PSF treatment in the past 10 years. Cauterization of the internal opening of the fistula yields results comparable to those of open neck surgery in terms of effectiveness. On the other hand with laser excision, postoperative pain was minimal and no airway obstruction was observed. NGT was used to prevent food soiling of the excised area and to minimize pain that would have occurred if no NG tube was inserted.

Per- and postoperative empirical antibiotics are essential and must be directed towards covering normal bacterial flora of the pharynx [20].

## Conclusion

Branchial cysts are important differential diagnosis in neck cysts. The present case report is the first branchial cyst to be reported in the pyriform fossa and the second branchial anomalies to be excised transorally with CO<sub>2</sub> laser.

**Conflict of interest** None.

**Ethical standard** Not applicable.

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