

Bilateral Pharyngoceles (Branchial Cleft Anomalies?) and Endoscopic Surgical Considerations

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A case report of bilateral pharyngoceles without a history of elevated intrapharyngeal pressures is used to support the hypothesis that pharyngoceles may be an adult manifestation of an internal branchial sinus anomaly. The development of a pharyngocele from a branchial sinus origin would suggest a predictable relationship to the hypoglossal, glossopharyngeal, and superior laryngeal nerves, which may influence the choice of surgical approach (open versus endoscopic) and the counseling of patients who are considering surgical correction.

Key Words: branchial region, diverticulum, dysphagia, laryngocele, pharyngocele.

INTRODUCTION

Pharyngoceles are well-described but rare anomalies.¹ Often, pharyngoceles are found in association with excessive coughing or blowing (playing wind instruments), and they generally occur at the junction between the superior and middle, or middle and inferior, pharyngeal constrictor muscles.^{1,2} These locations differ from that of the more commonly known Zenker's diverticulum, which arises between the inferior pharyngeal constrictor and cricopharyngeal muscles, in an area of weakness called Killian's triangle. Killian-Jamieson's diverticulum is found between the oblique and transverse fibers of the cricopharyngeal muscle, whereas Laimer's diverticulum is found between the cricopharyngeal muscle and the most superior esophageal wall circular muscles.³ Pharyngoceles are also often confused with laryngoceles, as both protrude through the thyrohyoid membrane. Laryngoceles, however, develop when the lateral saccule or laryngeal ventricle enlarges or dilates as a result of increased glottic pressure.¹

We present here the case of a healthy middle-aged man with bilateral lateral pharyngoceles without any history of activities that would result in elevated intrapharyngeal pressures. We also discuss the possibility that a pharyngocele may be an adult manifestation of an internal branchial sinus anomaly and how this information may influence the choice of surgical approach (open versus endoscopic) and patient counseling of risks.

CASE REPORT

A healthy 49-year-old man came to our clinic with

a 2- to 3-year history of progressive dysphagia, principally to pills and some solid foods, that was felt to be localized to the area of the larynx. The sensation passed after he swallowed liquids, but occurred on a daily basis. He specifically denied any excessive coughing, aspiration, food regurgitation, pain, hoarseness, recurrent bronchitis or pneumonias, and chronic throat clearing. However, he did complain of mucus collection only upon waking in the mornings that cleared up within an hour. Notably, the patient denied any history of playing wind instruments or participating in any other activities that require elevated intrapharyngeal pressures.

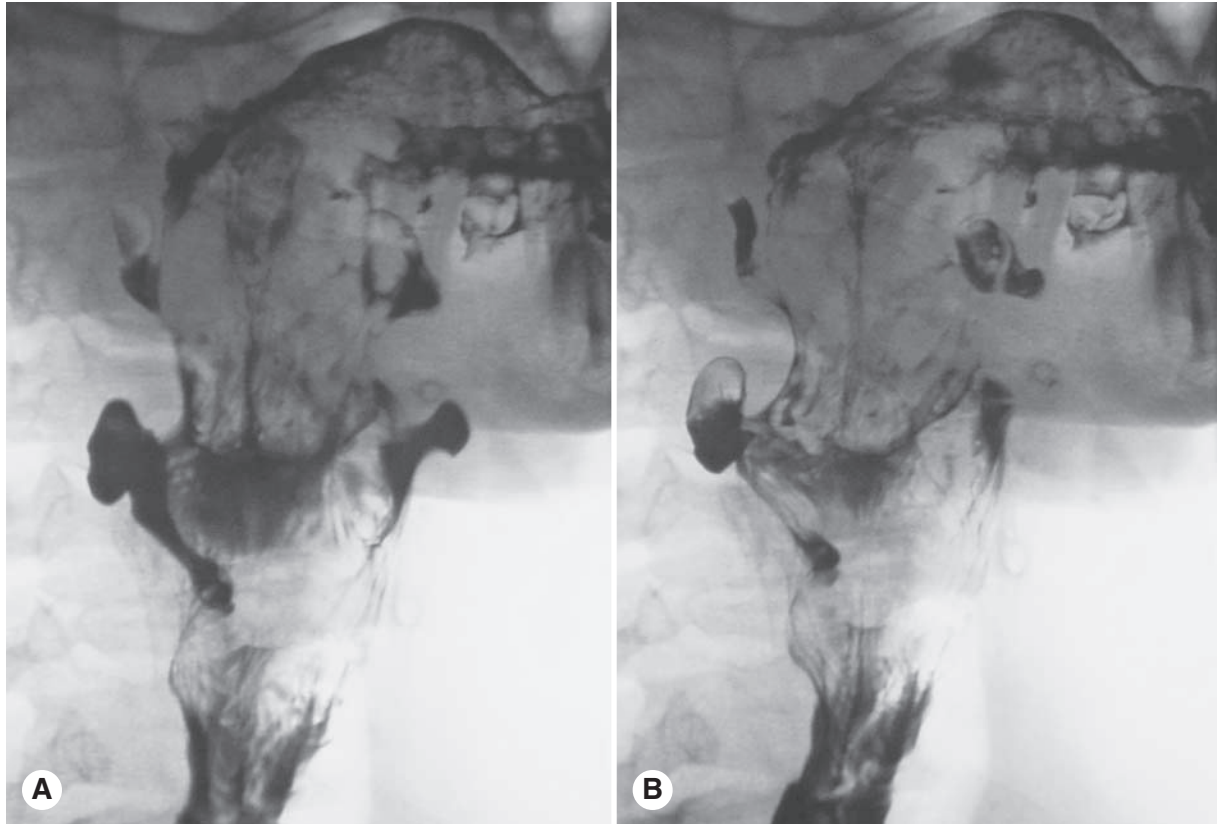
Our examination revealed normal findings, including normal vocal quality. Findings of fiberoptic nasopharyngolaryngoscopy were remarkable only for moderate posterior commissure hypertrophy consistent with laryngopharyngeal reflux. A barium swallow study revealed bilateral pharyngoceles, as well as deep pyriform fossae (see Figure). The patient declined any surgical intervention, and his symptoms were addressed through therapeutic swallowing maneuvers.

DISCUSSION

The first documented report of a pharyngocele was in 1886.² Since then, pharyngoceles have been rarely reported in the literature, but may occur more commonly than documented, as they can be asymptomatic.^{1,2,4-7} They are more common in men (between 3:1 and 8:1) and arise during the fifth and sixth decades of life.^{1,2} Their pathogenesis has been associated with muscle weakening with age, prior surgery,

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Barium swallow study. **A)** Bilateral pharyngoceles; one on right is larger than one on left. **B)** Image recorded moment later shows only right pharyngocele.

and increased intrapharyngeal pressure.² Along with these causes, we propose another possible explanation for the development of a pharyngocele. Although Norris² in 1979 mentioned in passing the confusion in distinguishing between pharyngoceles and embryological pouches, we suggest that some pharyngoceles may actually be yet another manifestation of a branchial arch anomaly — specifically, a small internal branchial sinus outpouching that dilates over time, resulting in a pharyngocele.² Indeed, branchial anomalies are known to present with a wide range of characteristics including, but not limited to, cysts, fistulas, and sinuses of the head and neck region. Second branchial arch anomalies constitute 95% of congenital branchial arch anomalies, with the majority of the rest derived from first and third branchial arch anomalies.⁸⁻¹⁰ Until recently, the existence of fourth branchial arch anomalies was doubtful, but they are now known to exist extremely rarely and are diagnosed almost exclusively in children with recurrent neck abscesses.⁸ Even more rarely are they initially discovered in adults.¹¹ Internal branchial sinus anomalies are hypothesized to be vestigial branchial pouches due to incomplete closure or failure of obliteration of the cervical sinus of His.⁹

Differentiation among the branchial anomalies on

clinical grounds alone is very difficult. Anatomically, an internal second branchial sinus would open near the tonsillar fossa and travel superior to the glossopharyngeal nerve, but below the stylohyoid ligament, lateral to the internal jugular vein at the level of the carotid bifurcation, extending between the internal and external carotid arteries. An internal third branchial sinus would open from the pyriform sinus and pierce the posterolateral aspect of the thyrohyoid membrane, remaining superior to the superior laryngeal and hypoglossal nerves but deep to the common carotid and/or internal carotid artery, as well as the glossopharyngeal nerve. However, an internal fourth branchial sinus would open from the pyriform fossa apex and pass inferior to the superior laryngeal nerve, thyroid cartilage, and cricothyroid muscle (fourth arch structures).^{8,10,12} The sinus may then loop around the aortic arch medial to the ligamentum arteriosum on the left, or around the subclavian artery on the right, before ascending posterior to the common carotid artery. Interestingly, fourth branchial pouch sinuses almost exclusively occur on the left side.⁸

Pharyngoceles almost always arise on the internal lateral aspects of the pharynx, just as do internal openings of branchial cleft sinuses.⁷ Pharyngoceles that

arise between the superior and middle pharyngeal constrictor muscles occur in an area inferior to the lower tonsillar pole on the lateral side of the vallecula.² This region corresponds to the site at which the glossopharyngeal nerve and stylohyoid ligament appear between the constrictor muscles and the site at which one would expect an internal second branchial sinus opening to be. Pharyngoceles that arise between the middle and inferior pharyngeal constrictor muscles occur in an area corresponding to the pyriform sinus. The region matches the site at which one would expect an internal third or fourth branchial sinus opening to be.

If a pharyngocele has a congenital branchial sinus origin, why do some cases remain asymptomatic into adulthood, when most cases of branchial sinuses present with recurrent infections in childhood? Just as there exist various degrees of branchial cleft anomalies such as preauricular pits and dimples, internal sinuses may initially exist as mere dimples or pits of various sizes, producing areas of anatomic weakness on the lateral pharyngeal mucosal surface. These initial branchial sinus manifestations may not be big enough to cause any food retention or problems in childhood, but over time, through repetitive deglutition, extrinsic pressures are exerted on the internal branchial sinus opening from swallowed foods propelled downward by the swallowing mechanism. Indeed, intrapharyngeal pressures up to 60 mm Hg are developed in the young and 20 mm Hg in the elderly.² Such pressures would gradually dilate the opening and enlarge the branchial sinus to the degree that it may become symptomatic or be discovered incidentally in adulthood and be (mis)diagnosed as a lateral hypopharyngeal diverticulum, pharyngocele, or laryngocele, depending on the location.

A branchial sinus origin for pharyngoceles would apply only in some cases. Unfortunately, to precisely define which among the above-mentioned diagnoses applies to a given presentation is often very difficult. Anatomic localization, embryonic knowledge, radiologic studies, and clinical presentation would all assist in coming up with the proper diagnosis, but in the end, precise anatomic dissection may be required for final determination — an intervention that is not always possible. In this particular case reported, the patient has a bilateral pharyngocele. Or could it be that it is an internal second branchial sinus (given its location) that has dilated over time, achieving its current appearance?

Is such differentiation even clinically relevant? Regardless of the precise diagnosis and pathogenesis, the nonsurgical treatment remains the same for all cases and may entail only external swallowing maneuvers, watchful monitoring, and antireflux medications until symptoms such as aspiration, regurgitation, and/or recurrent infections progress to the point at which surgical intervention is required. The surgical intervention would be excision by an external approach (or more recently, division by an endoscopic approach) of the common wall separating the pouch from the pharynx to create one confluent cavity — an easily performed outpatient procedure.^{5,13} The clinical relevance comes into play in considering the endoscopic approach, as there is a theoretical risk of injury to the hypoglossal, glossopharyngeal, and/or superior laryngeal nerves, given their proximity and relation to a branchial sinus anomaly. These nerves (as well as other potential neurovascular structures) are not identified at the time of common wall division. Theoretically, however, if a pharyngocele has a fourth branchial sinus origin, it should be safe for common wall division, as the sinus is inferior to these nerves. Likewise, a third branchial sinus origin would place the superior laryngeal and hypoglossal nerves at risk, but not the glossopharyngeal nerve. Last, a second branchial sinus origin would place all three nerves at theoretical risk.

Whether these theoretical concerns are clinically relevant has yet to be determined, given the limited endoscopic surgical experience in dealing with this rare finding in the patient population. However, an initial report suggests that good outcomes may be achieved, although further study in a greater number of patients is needed.⁵

CONCLUSIONS

If an endoscopic approach is considered to address a pharyngocele, it behooves the surgeon to cogitate on potential injury to the hypoglossal, glossopharyngeal, and/or superior laryngeal nerves. Exactly which nerves are at risk would depend on the pathogenesis and potential theoretical origin of a given pharyngocele. Unfortunately, precise presurgical, noninvasive determination is not easily performed with a high degree of certainty. Consideration that pharyngoceles may be branchial sinus anomalies provides a basis for counseling patients, although it must be kept in mind that not all pharyngoceles necessarily have an embryological origin.

REFERENCES

1. van de Ven PM, Schutte HK. The pharyngocele: infrequently encountered and easily misdiagnosed. *J Laryngol Otol* 1995;109:247-9.
2. Norris CW. Pharyngoceles of the hypopharynx. *Laryngoscope* 1979;89:1788-807.
3. Westrin KM, Ergun S, Carlsoo B, Zenker's diverticulum — a historical review and trends in therapy. *Acta Otolaryngol (Stockh)* 1996;116:351-60.

4. Chevalier P, Motamedi JP, Marcy PY, Foa C, Padovani B, Bruneton JN. Sonographic discovery of a pharyngocele. *J Clin Ultrasound* 2000;28:101-3.
5. Huang PC, Scher RL. Endoscopic management of lateral pharyngeal pouch. *Ann Otol Rhinol Laryngol* 1999;108:408-10.
6. Meehan T, Henein RR. An unusual pharyngeal pouch. *J Laryngol Otol* 1992;106:1002-3.
7. Restrepo S, Villamil MA, Rojas IC, Palacios E. Pharyngocele: CT and MRI findings. *Ear Nose Throat J* 2003;82:492, 494.
8. Godin MS, Kearns DB, Pransky SM, Seid AB, Wilson DB. Fourth branchial pouch sinus: principles of diagnosis and management. *Laryngoscope* 1990;100:174-8.
9. Choi SS, Zalzal GH. Branchial anomalies: a review of 52 cases. *Laryngoscope* 1995;105:909-13.
10. Ostfeld E, Segal J, Auslander L, Rabinson S. Fourth pharyngeal pouch sinus. *Laryngoscope* 1985;95:1114-7.
11. Lin CJ, Lin YS, Kang BH, Lee JC. Silent internal sinus of the pyriform fossa: a rare adult manifestation of a branchial anomaly. *Arch Otolaryngol Head Neck Surg* 2003;129:356-8.
12. Liston SL. Fourth branchial fistula. *Otolaryngol Head Neck Surg* 1981;89:520-2.
13. Chang CY, Payyapilli RJ, Scher RL. Endoscopic staple diverticulostomy for Zenker's diverticulum: review of literature and experience in 159 consecutive patients. *Laryngoscope* 2003;113:957-65.