Endoscopic Cauterization of Congenital Pyriform Fossa Sinus Tracts
An 18-Year Experience

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Branchial arch abnormalities opening into the pyriform fossa are an important congenital cause of recurrent neck abscesses in children. The classification and naming of these anomalies is controversial because of the complex embryological development of the region involved. In 1972, Sandborn and Shafer first described a neck mass caused by a sinus tract traveling from the left pyriform fossa to the superior pole of the left lobe of the thyroid. The lesion was classified as a fourth branchial pouch derivative, which became the widely used term for such structures. Theoretically, a complete fourth branchial pouch fistula tract would begin at the pyriform fossa, exit the larynx near the cricothyroid joint, and pass between the superior and recurrent laryngeal nerves behind the body of the thyroid gland. The fistula would then make a convoluted journey toward the mediastinum before looping back and exiting anterior to the sternocleidomastoid muscle on the lower neck. To our knowledge, a complete fistula tract naturally opening onto the neck has never been reported. To complicate matters, fistula tracts arising from the third branchial pouch theoretically would also start at the pyriform fossa. However, the tract would pass cranial instead of caudal to the superior laryngeal nerve. Because of the difficulty of distinguishing third and fourth pouch tracts, some authors have chosen to consider them a single entity. More recently, the thymopharyngeal duct, a third pouch derivative independent of the third and fourth fistula tracts, has been alternatively proposed as the cause of these lesions. The thymopharyngeal duct is formed when the thymus descends from the third pouch through the fourth arch to fuse with its contralateral counterpart during fetal development. Failure of thymopharyngeal duct involution should lead to a sinus tract containing thymus-derived tissue starting at the pyriform fossa and descending in close association with the thyroid.

IMPORTANCE Congenital pyriform fossa sinus tracts predispose to neck masses and neck abscesses in pediatric and occasionally adult patients. Traditional management involves open excision with substantial potential morbidity. Endoscopic management allows an alternative, less morbid treatment approach.

OBJECTIVE To evaluate the long-term effectiveness of endoscopic cauterization as definitive treatment for pyriform fossa sinus tracts.

DESIGN, SETTING, AND PATIENTS Retrospective review of the medical records of 23 children (aged 7 months to 14 years) with pyriform fossa sinus tracts treated with endoscopic cauterization between 1995 and 2013 at a tertiary care children’s hospital.

INTERVENTION Endoscopic electrocauterization of pyriform fossa sinus tract opening.

MAIN OUTCOMES AND MEASURES Recurrence of symptoms after endoscopic treatment.

RESULTS Twenty-one of 23 patients experienced no recurrence after their first endoscopic electrocauterization of the sinus tract. The 2 patients with recurrence experienced symptoms within 1 month of cauterization and were treated with either open excision or recauterization. Endoscopic cauterization was able to definitively treat 9 patients whose treatments with incision and drainage or open excision had failed. Mean (range) follow-up for the 15 patients with follow-up was 7.4 (0.10-14.2) years. No procedure-related morbidity was reported.

CONCLUSIONS AND RELEVANCE Endoscopic cauterization seems to be an effective and potentially permanent treatment for congenital pyriform fossa sinus tracts.
Because determining the origin of a sinus tract requires complete surgical excision of the tract, their exact classification is not possible in cases that do not involve neck dissection. Furthermore, the embryological cause of these branchial abnormalities is ultimately unimportant from a clinical perspective. Therefore, we refer to these lesions by their physical characteristics as pyriform fossa sinus tracts (PFSTs).

Since their first description, PFSTs have generally been regarded as very rare entities.4 However, a 2008 literature review found 526 published cases of PFST, 5-fold more than previously thought to exist.3 Many more cases have been reported since then, although PFST prevalence has varied in reports from a number of individual institutions.6-8 The sinus tracts occur equally in both sexes and overwhelmingly on the left side of the neck (94%).5 Usually, PFST presents in young children as either acute supplicative thyroiditis or recurrent neck abscess. Less commonly, neonates may present with a noninfective, compressible neck mass.9 The sinus tracts often remain undiagnosed for several years, and patients may undergo many unsuccessful abscess drainage procedures before appropriate treatment is obtained.3-4,10 Computed tomography (CT) or magnetic resonance imaging (MRI) showing an inflammatory neck mass closely associated with the thyroid can aid in diagnosis and determining the extent of the lesion. Direct laryngoscopy of the pyriform fossa plays an integral role in conclusive diagnosis and should always be conducted when a PFST is suspected.4,5,10

Pyriform fossa sinus tracts can be treated by means of either traditional complete excision with possible accompanying partial thyroidectomy or endoscopic cauterization of the pyriform fossa. We first proposed endoscopic electrocautery as a definitive treatment for PFST in 1998 and subsequently reported on 10 cases involving the procedure in 2004.4,11 The procedure is described in detail in our previous reports.4,11 With continued clinical experience since our initial report, we have found that cauterization alone without balloon dilation of the sinus tract seems to be equally efficacious (Figure 1). We present herein long-term follow-up information on the original case series, as well as 13 additional cases of patients with PFST who have presented to our institution.

Methods

We conducted a retrospective case review of 23 patients who received a diagnosis of PFST on the basis of CT and endoscopic findings between 1995 and 2013 at Children’s Medical Center of Dallas in Dallas, Texas. Only patients with PFST who received endoscopic treatment at some point in their care were included in our study. We also contacted patients by telephone to obtain long-term follow-up information. The study was conducted with approval from the institutional review board of the University of Texas–Southwestern Medical Center. Informed consent was waived because of the retrospective nature of the study.

Results

Sixteen girls and 7 boys with a mean age at presentation of 5.9 years were included in our study. Their clinical characteristics are summarized in the Table.

Clinical presentation of the 23 affected children included initially treated neck abscess (8 [35%]), recurrent neck abscesses (13 [57%]), supplicative thyroiditis (1 [4%]), and recurrent supplicative thyroiditis (1 [4%]). Two patients (9%) also presented with acquired hypothyroidism. All lesions occurred on the left side. Ten patients (43%) had a history of drainage attempts performed elsewhere that were unsuccessful in definitively treating the condition before diagnosis was made at our institution. We also preoperatively drained lesions on 13 patients (57%) to resolve the neck infection in anticipation of endoscopic treatment 4 to 6 weeks later. Two patients (9%) presented with recurrence after open excision of the lesion. Twenty-two patients (96%) underwent CT, and 2 (9%) underwent MRI. Computed tomographic scans in 10 cases (43%) were interpreted by the attending radiologist as suspicious for a PFST or equivalent structure. After PFST was suspected, a sinus tract opening was identified in all patients and usually during the first endoscopic examinations (mean [range], 1.2 [1-3] examinations) (Figure 2). After PFST was suspected, a sinus tract opening was identified in all patients and usually during the first endoscopic examinations (mean [range], 1.2 [1-3] examinations) (Figure 3).

We primarily treated 22 of the 23 patients using only the endoscopic approach. The remaining patient, patient 6 (Table),
was primarily treated with simultaneous endoscopic cauterization and open excision of the lesion and did not experience recurrence. Twenty of the 22 patients (91%) who received endoscopic cauterization alone were definitively treated without clinical recurrence after the first attempt. One patient, patient 18, required 3 more procedures for definitive treatment. This patient developed a recurrent neck infection within 1 month of the initial cauterization. The opening in the pyriform fossa was relocated and recauterized on examination. After the second recurrence, the sinus tract opening could no longer be located and the lesion was partially excised from the skin surface. After the third recurrence, complete external excision of the lesion with partial thyroidectomy was required for final treatment, and the patient has been free from recurrence for 6.6 years. Patient 22 experienced neck swelling within 2 weeks of initial cauterization that resolved with antibiotic therapy. On reinspection of the pyriform fossa, a small mucosal “dimple” was identified and recauterized at the site of the previous tract, although no obvious patent sinus tract opening was seen (Figure 4). The patient was problem free at 1-month follow-up. Our clinical suspicion is that residual cyst contents caused an initial inflammatory response after tract cauterization and that the cyst cavity has since scarred in.

The mean time from first presentation at our institution to final curative treatment was 7.4 months (median [range], 2.0 [0.0-68] months). No postoperative complications were reported in any patients. Patient 19 had a 2-year history of hypothyroidism concurrent with thyroiditis that resolved following cauterization of the PFST. Fifteen of 23 patients (65%) received follow-up in clinic. Eight of these patients were also more recently contacted by telephone. None of the patients expressed any additional neck complaints when contacted in follow-up. Among patients with follow-up, the mean duration without recurrence after final treatment was 7.40 years (median [range], 9.11 [0.1-14.2] years; n = 15). Of the 8 patients who did not follow up in clinic and who were not available for follow-up by telephone, none have presented again at our institution with complaints relating to PFST.

### Discussion

Knowledge and treatment of PFSTs have evolved over the last several decades. As with most disease processes, less invasive procedures are developed over time to reduce morbidity, but their success is always questioned at the outset. Our case
series contains some of the longest follow-up data on endoscopic cauterization of PFSTs and is, to our knowledge, the largest series on electrocauterization in the literature. This includes 6 patients who have reached adulthood without recurrence of neck infection since receiving treatment as school-aged children. These results suggest that the risk of PFST recurrence is minimal if the patient has been problem free for several years after closure of the sinus.

The 91% success rate after initial cauterization that we experienced is higher than has been reported elsewhere. Chen et al treated 9 patients in the same manner, without recurrence in 7 (78%). Using mainly laser cautery, Leboulanger et al treated 19 patients, of whom 13 (68%) were without recurrence after a mean of 23 months of follow-up. The latter group conducted follow-up endoscopy on all patients and treated 3 cases of asymptomatic reopening of the sinus tract, which may explain their lower reported success rate. Cha et al alternatively used trichloroacetic acid chemocauterization in 44 patients with an initial success rate of 77%. Compared with other forms of cautery, electrocauterization may be superior in permanently closing the sinus tract. Electrocauterization also seems to be comparable in efficacy to the traditional approach of open excision, which has a reported success rate of 85% overall and 92% when performed with partial thyroidectomy. Notably, our results include 2 patients (Table) who were treated endoscopically after unsuccessful open ex-
cisions. In 1 case, cauterization cured the patient after 3 failed attempts at excision.

Postoperative complications, most commonly vocal fold paralysis, have been reported in a small number of cases involving open excision. Superior and/or recurrent laryngeal nerve injury with resultant vocal cord paresis is a theoretical complication of endoscopic cauterization. None of our patients had vocal complaints or signs or symptoms of paresis. Postoperative recovery is smooth, without substantial dysphagia or odynophagia. In fact, to our knowledge, no complications have ever been reported from endoscopic cauterization.

Our single patient with multiple recurrences did not improve after a second endoscopic attempt and developed a neck infection even after the sinus opening was demonstrably eliminated. Cha et al proposed a reasonable protocol that reserves open excision for cases that persist after 3 attempts at endoscopic management. After multiple recurrences, we recommend a combined external and endoscopic approach. Excision with possible partial thyroidectomy should be performed alongside endoscopy to ensure that the entire sinus tract is removed and that the sinus has not reopened. Although some authors have suggested that the combined approach should be used initially as the standard for treatment, we believe it best reserved for cases that persist after endoscopic management alone. In a case series by Pahlavan et al, 1 patient experienced treatment failure with both cauterization and excision with hemithyroidectomy. This patient was eventually treated by means of pharyngotomy with obliteration of the left pyriform fossa.

The patients in the present study and those in other reports often have a long history of unsuccessful neck drainage procedures before the correct diagnosis is made. Although incision and drainage may be indicated for patients with acute infection that is not responsive to antibiotic treatment alone, it does not definitively address the underlying congenital anomaly and in fact may lead to fistula formation. We therefore continue to advocate for examination of the pyriform fossa in children with neck abscesses or fluid collections in the region of the third and fourth branchial apparatus, especially those that are recurrent or occur on the left side, so that a more definitive treatment option may be identified. Although we did not discover any symptomatic or asymptomatic lesions of the right side of the neck in our series, should an asymptomatic right-sided PFST be discovered during examination, we would likely proceed with cauterization because the risk of adverse effects is low.

In some cases, the sinus opening may not be found during the first examination. Suspension microlaryngoscopy with manipulation of the mucosa of the pyriform sinus (Figure 3) has been helpful in identifying the sinus tract opening, especially in the nonacute setting. If an opening is not initially identified, localization should be reattempted after inflammation in the area has been controlled medically. In addition, in the acute setting, compression of the neck mass during laryngoscopy to force purulent emissions into the pyriform fossa can help locate an elusive sinus opening or at least confirm its existence. In our experience, CT scan or MRI may be useful in raising suspicion for the diagnosis, but these techniques cannot specifically locate the sinus tract opening. Contrast swallow studies have been used to confirm the presence of a PFST and perhaps more specifically locate the tract. Whereas a meta-analysis indicates that contrast swallow studies have positive predictive values for the identification of sinus tracts that are similar to those of endoscopy (88% vs 90%, respectively), we have not routinely used them in the diagnosis and management of such lesions because endoscopy affords the additional benefit of enabling immediate and potentially definitive management.

The terminology of PFST itself has evolved since we last reported on the subject. The term “fourth branchial pouch cyst” and its various permutations, which have traditionally been used to describe these anomalies, have lately fallen out of favor because of a lack of consensus about their origin. Echoing statements made by other authors, we believe that the term “pyriform fossa sinus tract” best describes the important diagnostic feature of the abnormality and suggest its use to avoid confusion in the literature. Without neck dissection and definitive mapping of the tract, other nomenclature is merely speculative regarding the origin of the lesion. Furthermore, as noted by Thomas et al, defining these lesions as a “sinus” rather than cyst or fistula is important because a
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Original Investigation Research

Conclusions

Regardless of their embryologic origin, PFSTs have become increasingly reported in recent years. This emphasizes the importance of their consideration by clinicians dealing with recurrent neck infections, especially those located on the left side of the neck. We have presented 1 of the largest series of cases of PFST with the longest follow-up on PFST cauteralization in the literature to date. The results reaffirm our previous recommendation of using endoscopic electrocauterization as the first line of treatment for these abnormalities. The approach was successful in most of our patients after the first treatment, and the results have been maintained over time. As with open excision, recurrence can occur. Nevertheless, we believe that the endoscopic approach provides substantial benefits as a fast, minimally invasive procedure that decreases morbidity while providing long-term treatment efficacy comparable to that of traditional open techniques.