Systematic Review of Endoscopic Obliteration Techniques for Managing Congenital Piriform Fossa Sinus Tracts in Children

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Abstract

Background. Piriform fossa sinus tracts (PFSTs) are a recognized cause of recurrent deep neck infections in the pediatric population. Conventional management has historically required open resection, but over recent years minimally invasive endoscopic approaches to obliterate the pharyngeal opening of the sinus have been performed in many centers. However, there is a lack of clear evidence regarding the success rate and safety of these approaches.

Objective. To determine the success rate of endoscopic management of PFST through a systematic review of the existing literature.

Data Sources. MEDLINE (1964-2014) and bibliographies of identified papers.

Review Methods. Two authors independently reviewed 170 abstracts and identified relevant studies for full-text review. Data were independently extracted from those studies, and the Oxford Centre for Evidence-Based Medicine guidelines were used to classify the level of evidence.

Results. Thirteen studies met the inclusion criteria, comprising a total of 84 patients. All included studies were evidence level 4 (case series). Various methods of obliterating the PFST were described: electrocautery (n = 39), laser (n = 19), trichloroacetic acid (n = 19), silver nitrate (n = 4), combination of silver nitrate and laser (n = 2), and fibrin glue (n = 1). The success rate for endoscopic management of PFST was 89.3% overall (90.5% in primary cases and 85.7% in revision cases). The only adverse event reported was temporary vocal cord immobility in 2.4% (n = 2) of cases.

Conclusion. Endoscopic management of pediatric PFST appears to be safe and effective, as a primary option and for revision after open surgery.

Keywords

piriform fossa sinus tract, branchial cleft anomaly, endoscopy, cauterization

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Background

A piriform fossa sinus tract (PFST) is a rare congenital anomaly in which there is an abnormal communication between the mucosa of the piriform fossa and the neck. It usually presents as recurrent deep neck infections or suppurative thyroiditis, as the tract often ends in the thyroid gland. It presents most commonly in the pediatric population and is overwhelmingly more prevalent on the left than on the right.1,2 A child presenting with an abscess in the lower lateral spaces of the neck or with suppurative thyroiditis is a clinical scenario that should prompt the otolaryngologist to perform a direct laryngoscopy in the operating room. With careful examination of the piriform fossa, the opening of a tract can be identified in many of these patients, confirming the diagnosis of a PFST. These anomalies have also been known as third or fourth branchial cleft anomalies. It has been suggested, however, that they are in fact related to failure of the obliteration of the thymopharyngeal duct3 and are now widely referred to as PFST (see Figure 1). These anomalies have historically been managed predominantly by open resection of the tract with or without hemithyroidectomy.2,4 With time, it became apparent that recognizing and addressing the internal opening helped in successful open surgical management and guiding the dissection.1 Cannulating the opening and ligating it at the piriform fossa apex is advocated when open

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Resection is chosen. In more recent years, there has been growing interest in managing PFSTs through endoscopic obliteration or cauterization of the tract as a primary treatment, therefore avoiding the need for an open procedure altogether. A number of methods for minimally invasive endoscopic tract obliteration have now been described, but to date there has not been a systematic appraisal of the efficacy and risk of adverse events of these techniques and approaches.

We therefore conducted a systematic review of the literature identifying and appraising the evidence for endoscopic obliteration techniques in the management of congenital piriform sinus tracts in children. This review aims to assist otolaryngologists in the management of this congenital anomaly. Specifically, it has the potential to help clinicians in deciding whether to consider an endoscopic technique and an estimate for the anticipated success rate in primary and revision cases. Additionally, the review explores any adjuvant techniques that may influence the success rate when an endoscopic approach is chosen.

Methods

Search Strategy

The MEDLINE database was searched for any relevant paper published between 1946 (creation of MEDLINE) and May 2014 about endoscopic management of PFST. A detailed search strategy was developed with the following search terms: “piriform fossa sinus tract” OR “third branchial cleft sinus” OR “fourth branchial cleft sinus” OR “third branchial cleft anomaly” OR “fourth branchial cleft anomaly” OR “piriform sinus tract” AND “endoscopic management” OR “endoscopic cauterization” OR “endoscopic obliteration” OR “cauterization.” Each identified abstract was evaluated independently by 2 reviewers.

Criteria for Inclusion

The eligible studies were randomized controlled trials, observational studies (cohort studies, case-control studies), and case series of pediatric patients diagnosed with a PFST and undergoing endoscopic management. Studies written in French or English were included. Studies looking exclusively at an adult population or using a combined open and endoscopic approach were excluded. Any disagreement between the reviewers was resolved by discussion. The bibliography of each included study was scanned for any more articles potentially meeting our inclusion criteria (see Figure 2).

Assessment of Quality

The included studies were graded according to the Oxford Centre for Evidence-Based Medicine levels of evidence. Because this systematic review includes many case reports and case series (a rare condition), an additional system of quality assessment was used to minimize biases. The risk of selection, performance, detection, attrition, and reporting bias in case series was assessed by determining a score from 0 (low risk) to 5 (high risk) according to the following system:

1: Sample selection: consecutive or not; 1 = no or not stated, 0 = consecutive
2: Diagnostic criteria stated: 1 = not stated, 0 = stated
3: Outcomes measured consistently for all patients: 1 = not consistent, 0 = consistent
4: Outcomes reported consistently for all patients: 1 = not consistent, 0 = consistent
5: Follow-up period ≥1 year: 1 = <1 year, 0 = ≥1 year

This allowed a more detailed view of the quality of included papers. The specific biases that are being appraised for risk are as follows: selection bias (1 and 2), classification bias (2 and 3), reporting bias (3, 4, and 5), and recall bias (5). We designed this system based on a similar system used in a previous systematic review. The principles of this scoring method were based on the Cochrane Handbook for Systematic Reviews of Interventions.
Data were extracted from the identified studies, including the number of patients, patient demographics, diagnostic methods, intervention, adjuvant techniques, outcome measure, and complications or adverse events. Success rates were determined for primary and revision cases, as well as for techniques and per number of treatments.

The study did not need approval by the University of British Columbia / Children’s and Women’s Health Center Research Ethics Board, as it did not involve any patients or patient records.

Results

Our initial search resulted in 170 abstracts. Twenty-one of these were considered for further review, and 3 more were found by reviewing the reference lists of the selected papers. After full-text readings, 13 of these 24 papers were eligible for inclusion in this review. Table 1 shows the included studies with information on the number of patients, technique, and success rate. Of the exclusions, 4 evaluated open procedures for PFST; 3 were about the wrong topic; 2 included duplicated data; 1 referred to an adult case; and 1 did not provide any information about the ages of the subjects and we therefore could not include it in this strictly pediatric review. Attempts were made to contact the authors to obtain the missing information but were unsuccessful. All included studies were case series (Oxford Centre for Evidence-Based Medicine level of evidence 4).

Results of our risk-of-bias quality assessment are showed in Table 2. We obtained a median score of 1 (out of 5), suggesting that the risk of bias was low.

The included papers yielded a total of 63 pediatric patients primarily treated endoscopically for a PFST. An additional 21 patients in these studies had been treated after having had an open procedure as an attempt to resect the fistula prior to the endoscopic approach, and their data were analyzed separately as revision cases. We did not consider a simple incision and drainage as an open procedure and still included the patients who had had an incision and drainage previous to or together with the endoscopic treatment in the primary group. There were 31 females and 13 males in the primary group (information was missing for 19 patients) and 16 females and 5 males in the revision group. The average age was 6.9 years in the primarily treated patients and 7.1 years in the revision group. Average follow-up was calculated for the 54 cases for which the information was available: 50.4 months in the primary group (range, 4 months to 15.4 years) and 19.7 months in the revision group (range, 2 to 98.4 months).

Techniques Used

Electrocautery was the most commonly used technique for obliterating the PFST, used in 46.4% of the identified cases (39 patients). Other techniques were laser (n = 19, 22.6%), trichloroacetic acid (n = 19, 22.6%), silver nitrate (n = 4, 4.8%), combination of silver nitrate and laser (n = 2, 2.4%), and fibrin glue (n = 1, 1.2%).

There were several different lasers used, including $\text{CO}_2$-diode (Ceralas D), and Thulium laser.

Few studies mentioned any adjuvant techniques or routine postoperative care. We therefore were unable to

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Figure 2. Flowchart of search strategy and included studies.
determine if any adjuvant technique or routine care practice had any influence on the success rate of the procedure.

Success Rate for Primary Treatment

For the 63 patients treated endoscopically as a first attempt, success rate was 77.8% (49 of 63) after 1 treatment, 87.3% (55 of 63) after 2 treatments, and 90.5% (57 of 63) after 3 treatments. No patient had undergone 3 attempts.

Success Rate for Revision Cases

For the 21 patients in the revision group, the success rate was 71.4% (15 of 21) after 1 attempt and 85.7% (18 of 21) after 2 attempts. No patient had undergone ≥2 treatments. Overall, the success rate is 89.3%, with 75 of 84 reported patients with no recurrence after endoscopic management of a PFST. In most studies, success was defined as no recurrence of symptoms or neck infection, but a few studies did confirm obliteration with a barium swallow or direct exam.9,12-15

Success Rate per Technique

Electrocautery obliteration had a success rate of 92.3% (36 of 39 patients). Laser was successful in 84.2% (16 of 19). Trichloroacetic acid had a success rate of 78.9% (15 of 19); silver nitrate alone and combined with laser as well as fibrin glue all had 100% success rate but included only a small number of patients (4, 2, and 1 patients, respectively).

Table 1. Included Studies with Technique Used and Reported Success Rate.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Patients, n</th>
<th>Technique</th>
<th>Success Rate, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ahmed</td>
<td>2008</td>
<td>3 revision</td>
<td>Electrocautery</td>
<td>100</td>
</tr>
<tr>
<td>Cigliano</td>
<td>2004</td>
<td>1 primary</td>
<td>Fibrin glue injection</td>
<td>100</td>
</tr>
<tr>
<td>Chen</td>
<td>2009</td>
<td>7 primary, 2 revision</td>
<td>Electrocautery, plus sutures in 7 cases</td>
<td>77.8</td>
</tr>
<tr>
<td>Kim</td>
<td>2000</td>
<td>1 primary, 10 revision</td>
<td>TCA, 10%-40%</td>
<td>81.8</td>
</tr>
<tr>
<td>Leboulanger</td>
<td>2010</td>
<td>19 primary</td>
<td>CO2 laser (n = 13), Thulium laser (n = 4), and electrocautery (n = 2)</td>
<td>84.2</td>
</tr>
<tr>
<td>Miyauchi</td>
<td>2009</td>
<td>4 primary, 1 revision</td>
<td>TCA, 30% with multiple 1-minute applications</td>
<td>80</td>
</tr>
<tr>
<td>Park</td>
<td>2012</td>
<td>2 primary</td>
<td>TCA, 30%</td>
<td>100</td>
</tr>
<tr>
<td>Pereira</td>
<td>2008</td>
<td>1 primary, 1 revision</td>
<td>Silver nitrate</td>
<td>100</td>
</tr>
<tr>
<td>Sayadi</td>
<td>2006</td>
<td>1 primary, 1 revision</td>
<td>CERALAS D laser, 980 nm</td>
<td>100</td>
</tr>
<tr>
<td>Stenquist</td>
<td>2003</td>
<td>1 primary</td>
<td>TCA, 40%</td>
<td>100</td>
</tr>
<tr>
<td>Sun</td>
<td>2014</td>
<td>20 primary, 2 revision</td>
<td>Electrocautery</td>
<td>95.5</td>
</tr>
<tr>
<td>Watson</td>
<td>2013</td>
<td>5 primary</td>
<td>Electrocautery (n = 1), CO2 laser plus silver nitrate (n = 2), silver nitrate (n = 2)</td>
<td>100</td>
</tr>
<tr>
<td>Wong</td>
<td>2014</td>
<td>1 primary, 1 revision</td>
<td>Electrocautery</td>
<td>100</td>
</tr>
</tbody>
</table>

Abbreviation: TCA, trichloroacetic acid.
*Complications included temporary vocal cord immobility (n = 2).

Table 2. Risk of Bias: Quality Assessment Scoring of Included Studies.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Sample</th>
<th>Diagnosis Criteria</th>
<th>Outcome Measure</th>
<th>Outcome Reported</th>
<th>Follow-up</th>
<th>Total Score</th>
</tr>
</thead>
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<tr>
<td>Ahmed</td>
<td>2008</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Cigliano</td>
<td>2004</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>4</td>
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<tr>
<td>Chen</td>
<td>2009</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
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<tr>
<td>Kim</td>
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<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
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<tr>
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<tr>
<td>Miyauchi</td>
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<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
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<tr>
<td>Park</td>
<td>2012</td>
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<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Pereira</td>
<td>2008</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Sayadi</td>
<td>2006</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Stenquist</td>
<td>2003</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Sun</td>
<td>2014</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Watson</td>
<td>2013</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Wong</td>
<td>2014</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
</tbody>
</table>
Complications
The only complication reported was temporary vocal cord immobility, occurring in 2 cases (2.4%). Both reported incidents occurred after trichloroacetic acid cauterization.\textsuperscript{15}

Discussion
The diagnosis of a PFST, once suspected, is fairly straightforward. However, the method by which it is addressed is far less consistent. As endoscopic approaches are becoming more common, we can find various reports and case series in the literature. Through this review, the available data on this specific topic have been collected and analyzed. The study quality was overall good with a low risk of bias, as noted by our median score of 1 out of 5. However, the risk of a selection bias in particular has to be considered when reviewing the literature, as demonstrated by the included studies scoring more poorly on this than the other criteria in our quality assessment evaluation (see Table 2).

PFSTs, or third and fourth branchial cleft anomalies, have been recognized as a cause of recurrent deep neck abscesses and supplicative thyroiditis for a number of years. Once identified through barium swallow, computed tomography, magnetic resonance imaging, or direct laryngoscopy, the opening of the tract in the piriform fossa can be addressed. Several reports of open procedures for resection of PFST insist on the importance of addressing the internal opening to reduce recurrences. Open resection of PFST has a failure rate of about 15%.\textsuperscript{2,4} Moreover, these procedures have a 6% complication rate, the most common complications being vocal cord paralysis, salivary fistula, and infection.\textsuperscript{2} In comparison, we found a 2.4% complication rate in the reviewed literature for endoscopic procedures, all of which was temporary vocal cord immobility.

Endoscopic procedures to cauterize the opening of a PFST can be curative, therefore avoiding the risks of open procedures. Success rates between 78.9% and 100% were reported, depending on the technique used and the number of treatments performed. The most commonly used techniques were cautery with monopolar diathermy, laser, and trichloroacetic acid. We found success rates to be comparable when looking at all the techniques described and therefore conclude that there is no evidence to support the superiority of one technique over another. Further studies would be needed to adequately compare laser versus chemical versus electrocauterization techniques. Some of the reports described up to 3 separate treatments before obtaining complete closure of the internal opening of the sinus tract. As morbidity was very low, multiple attempts at endoscopic obliteration of a PFST can be considered in the case of recurrent symptoms. Consideration should be given to the potential for inflammation of tissues after multiple endoscopic cauterization procedures and the possibility that this might make subsequent open surgery, where needed, more challenging. However, the limited data available did not demonstrate these issues after up to 3 repeated procedures.

This review also evaluated the success of secondary endoscopic cauterization techniques in patients who had previously undergone an open procedure as a technique to resect the tract. We found the success rate to be comparable in these secondary cauterization cases to primary cauterization cases (85.7% vs 90.5%). In view of the smaller number of patients for review in this group (21 vs 63 for primary cases), it remains difficult to draw conclusions from these data. However, because this is a low-risk procedure, we believe that it can be reasonably considered a “rescue” technique in patients who have recurrent symptoms after an open resection.

Very few authors discussed treatment adjuncts or specific postoperative care in their reports. Chen et al\textsuperscript{17} used sutures in addition to cauterization in some of their patients, but no conclusion can be drawn as far as the usefulness or necessity of this, because of the small numbers reported. It seems that the procedure is most often performed as outpatient day-case surgery and that there were no special precautions taken about resuming a normal diet. Kim et al\textsuperscript{13} mentioned withholding oral feeds for 7 days in their first 2 patients and then changed to allowing them oral feeds from postoperative day 2 onward without problems. There was no apparent consensus on the routine use of perioperative antibiotics.

Conclusion
Endoscopic cauterization of a PFST is a minimally invasive technique with success rates that compared well to those of open surgery and with less morbidity. We believe, given the existing literature, that this is a safe and effective procedure to treat a newly diagnosed PFST. It can also be considered for treating a recurrence after open resection, but the available evidence for this is relatively limited. Described endoscopic ablative techniques include monopolar cautery, laser, and trichloroacetic acid application, with no evidence for superiority of one technique. Some treatment adjuncts have been described, such as application of a suture to the PFST opening, but to date no conclusion can be drawn about their potential for added efficacy. Repeated endoscopic treatment attempts can be performed to obtain complete closure of the opening of the tract if required. Due to the rarity of this condition, it is unlikely that it would be pragmatic to design a randomized trial. However, this review highlights some questions that could be addressed by multicenter prospective data collection or through a multicenter observational study, which may provide some support for the success of one cauterization technique over others.

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Sophie Lachance, design of the study question and search strategy, search, data extraction and analysis, drafting of manuscript, final approval of the version to be published and agreement to be accountable for all aspects of the work; Neil K. Chadha, supervision of the designing of the study question and search strategy,
Disclosures

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References