ORIGINAL RESEARCH

Revised: 28 October 2022

A safe and effective surgical method for complex pyriform sinus fistula

Yun Li M.M. | Huijun Yue M.D. | Guoqing Lei M.M. | Fanqin Wei M.D. | Zhangfeng Wang M.D. | Xiaolin Zhu M.D. | Wenbin Lei M.D. •

Otorhinolaryngology Hospital, The First Affiliated Hospital, Sun Yat-sen University, Guangzhou, Guangdong, People's Republic of China

Correspondence

Wenbin Lei, Otorhinolaryngology Hospital, The First Affiliated Hospital, Sun Yat-sen University, 58 Zhongshan Road II, Guangzhou, Guangdong, 510080, People's Republic of China. Email: leiwb@mail.sysu.edu.cn

Funding information

the 5010 Clinical Research Program of Sun Yat-sen University, Grant/Award Number: 2017004; the National Key R&D Program of China, Grant/Award Number: 2020YFC1316903; the General Program of National Natural Science Foundation of China, Grant/Award Number: 81972528

Abstract

Objective: This article aims to propose a new surgical method for the treatment of pyriform fistula, especially for the complex pyriform fistula.

Methods: A total of 36 patients with pyriform fistula underwent the procedure between August 2017 to October 2020. Surgery was performed by the senior authors using the same technique at the same clinical center for all patients. The median follow-up time was 33 months. Meantime, we collected information on patients with pyriform fistula using traditional surgical methods in our hospital from April 2015 to November 2018 for comparison.

Results: The surgery was successfully completed in 36 patients. In all, 32 patients had a history of multiple incisions and drainage, 16 patients had a history of surgical resections, and two patients had a history of cauterization of the internal fistula. Compared with traditional surgical methods, our new surgical method greatly shortens the length of the surgical incision (4.3 vs. 5.5, p < 0.0001), reduces the operation time (8.1 vs. 27.1, p < 0.0001), and reduces the blood loss (103.2 vs. 196.8, p < 0.0001). None of the 36 patients in this study had complications such as pharyngeal fistula, recurrent laryngeal nerve paralysis, or hypothyroidism. The mean follow-up duration after the excision of the lesion was 34.1 months. To date, no patients have relapsed.

Conclusion: Our experience showed that this surgical technique could be used to completely remove the fistula, and it was easier to perform than the conventional strategies. These treatment options result in less trauma and reliable results, especially for complex pyriform fistulas.

Level of evidence: IV

KEYWORDS

congenital neck mass, endoscopy, head and neck (benign), infectious/inflammatory, thyroiditis

Yun Li and Huijun Yue are co-first authors and contributed equally to this work.

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made. © 2022 The Authors. *Laryngoscope Investigative Otolaryngology* published by Wiley Periodicals LLC on behalf of The Triological Society.

1 | INTRODUCTION

Pyriform sinus fistula is a comparatively rare congenital disease of the neck. It originates from abnormal development of the third or fourth branchial clefts during embryonic development.¹⁻³ With the development of increasing knowledge about it, the incidence is on the rise and the treatment of the disease has attracted more and more attention. Complete resection of the fistula in the quiescent period of inflammation is still a clear way to cure the disease.^{4.5} However, this conventional surgery is challenging and results in large incisions with obvious postoperative scars. Complications and recurrence can also occur.^{6.7} These outcomes can greatly affect a patient's quality of life and even cause great discomfort.

In recent years, with the advancement of endoscopy technology, researchers have increasingly advocated for endoscopic cauterization of the sinus tract opening in the pyriform sinus.⁸⁻¹⁰ Unfortunately, this method leaves the fistula epithelium and creates a closed cavity in the closed pyriform sinus fistula, which increases the risk of recurrence. Moreover, relapse has been found to occur after a secondary infection or low resistance, especially when the fistula passes through the thyroid tissue.¹¹⁻¹³ Therefore, a better surgical method to treat this disease is needed.

The typical presentations of lesions in pyriform sinus fistula are recurrent low-neck abscesses, acute suppurative thyroiditis, and neck masses.^{14–16} Approximately, 90% of these sinus tracts are situated on the left side of the neck, which may be due to embryonic development.¹⁷⁻¹⁹ An internal fistula is usually located at the tip or base of the pyriform fossa.^{20,21} Generally, the pyriform sinus fistula passes through the larynx near the cricothyroid joint and then partially passes through or terminates at the upper level of the lateral lobe of the thyroid.^{14,22,23} Due to repeated infection and even rupture, the trajectory of the fistula may also extend to the base of the neck.^{24,25} For patients with complex pyriform fistulas. especially those whose fistula tissue is found to pass through the thyroid in preoperative evaluation, we proposed a surgical method of CO₂ laserassisted microsurgery combined with minimally invasive cervical surgery for the complete removal of pyriform sinus fistulas with minimal trauma. This surgical technique is easy to perform and we believe it could be used to completely remove the fistula with less trauma.

2 | MATERIALS AND METHODS

2.1 | Ethics approval and consent to participate

This was a retrospective case series and this study was approved by the Sun Yat-sen University Ethics Committee for Research and Publication.

2.2 | Patients

The study group comprised all eligible patients undergoing fistulectomy for pyriform sinus fistula at our institution during a three-year period (from August 2017 to October 2020). Meantime, we collected information on patients with pyriform sinus fistula using traditional surgical methods in our hospital from April 2015 to November 2018 for comparison. Age, sex, side of the presentation, mode of presentation, diagnostic investigations, treatment, operative details, and postoperative situation were noted from the case records. Complications and recurrence were also recorded at follow-up. Diagnoses were made based on clinical manifestation, preoperative examination, intraoperative findings, and postoperative pathology.

Patients with pyriform sinus fistula who met one or more of the following conditions were eligible: (1) patients with a thyroid abscess or preoperative evaluation showing that the fistula tissue passed through the thyroid; (2) patients who relapsed after their internal fistula was closed by chemical or physical cauterization; and (3) patients with a third or fourth branchial cleft cyst.

The exclusion criteria were as follows: (1) patients with active inflammation; (2) patients in whom the internal fistula of the pyriform fossa could not be fully exposed under suspension laryngoscopy; (3) patients with coagulation disorders; and (4) patients with other severe systemic diseases who could not tolerate surgery.

2.3 | Preoperative management

We encouraged the use of video laryngoscopy (KARL STORZ co., Tuttlingen, Germany) for the examination of patients in a supine position who performed a balloon-blowing action, which could better expose the bottom of the pyriform fossa. If it was still not well exposed, the operator could lift the larynx to assist with the observation (Figure 1). An enhanced scan of the fistula was obtained with thin-slice CT after oral administration of the contrast agent iopromide during the quiescent period of inflammation. Moreover, ultrasound was used to identify the presence of thyroiditis, which helped determine the relationship between the fistula and the thyroid. All eligible patients, including young children, underwent chest X-rays, electrocardiograms, and comprehensive laboratory tests, including routine blood tests, coagulation function tests, liver and kidney function tests, blood biochemical tests, and C-reactive protein tests to rule out surgical contraindications.

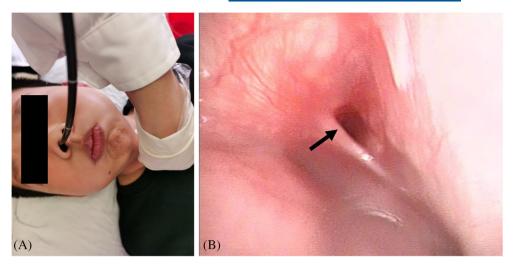
2.4 | Surgical equipment and instruments

The following devices were used: an S88 microscope (Oberkochen, Germany); a suspension laryngoscope and laryngeal microsurgical instruments (KARL STORZ co., Tuttlingen, Germany); a CO₂ laser (Lumenis AcuPulse 40 CO₂ laser, wavelength 10.6 μ m, Lumenis Ltd., Yokneam, Israel); and the Storz Professional Image Enhancement System (SPIES) (KARL STORZ co., Tuttlingen, Germany).

2.5 | Conventional surgery technique

The conventional surgeries were performed as traditional opening neck surgery according to the previous article.⁴ Briefly, we performed en bloc resection of the free lesion along the prevertebral fascia in a manner similar to selective neck dissection. After ligating and suturing

FIGURE 1 Electronic laryngoscope with balloon-blowing method. $(\rightarrow: fistula)$



the end of the branchial cleft deformities, we placed a silicone drainage tube in the surgical field. Finally, we inserted a nasogastric tube.

2.6 | New surgical technique (Video S1)

The surgeries were performed by the senior authors using the same technique and starting in 2018, this technique is routinely used in all eligible patients unless the patient cannot fully expose the pyriform fossa under suspension laryngoscopes and endoscopy. The procedure was performed with the patient in a supine position under general anesthesia with endotracheal intubation. Shoulder roll was used to lift the shoulders so that the neck was slightly extended. The patient was prepped and draped in the usual sterile fashion.

2.7 | Step 1 Internal pyriform sinus fistulectomy

We applied a suspension laryngoscope and endoscope to fully expose the internal fistula on the affected side of the pyriform fossa. Under the operating microscope, we used a carbon dioxide laser to make a circular incision around the mouth of the fistula (Figure 2A). The laser settings were superpulse, continuous wave mode, and 2-watt energy delivery. With the assistance of a micro-forcep, we performed stripping and separating along the longitudinal axis of the fistula against the tissue space outside the fistula (Figure 2B). Notably, the depth of the resection should not involve the muscle layer. When we dissected the fistula from front to back to reach the thyroid cartilage lamina, we cut off the fistula. Then, the residual fistula tissue was directly cut and vaporized by the laser (Figure 2C), which allowed us to maximize the removal of the lining epithelium of the fistula. After removing the fistula, we placed an absorbable gelatin sponge (0.2*1 cm) with a staining agent in the bottom of the surgical wound (Figure 2D), which was used as a marker for tracing and positioning the fistula in the following open surgery. Finally, we drew the margin of the pyriform fossa mucosa and closed the wound with eight-zero absorbable Prolene sutures in three-four stitches (Figure 2E).

After the internal fistulectomy was completed, we inserted a nasogastric tube, withdrew the endoscope, and switched to an external cervical approach for external fistula resection.

2.8 | Step 2 External pyriform sinus fistulectomy (similar to thyroid lobectomy or partial lobectomy)

The patient was positioned supine with an extended neck, and the cushions were removed. First, we made an approximately 4 cm horizontal incision along the neck skin lines at the level of the cricoid cartilage near the fistula. It is worth noting that we chose the original neck incision as frequently as possible. Second, we started the dissection from the boundary between the scar and normal tissue with a combination of blunt separation and sharp separation (Figure 3A,B). Then, we separated the fistula along its longitudinal axis and traced it to the "blue" mark (Figure 3D). Generally, the fistula is located below the thyroid cartilage on the affected side. If necessary, we also removed the lower corner of the thyroid cartilage and additional thyroid cartilage to fully expose the fistula, thereby facilitating its removal.

For patients with little inflammatory scar tissue in the neck and no evident inflammation in the thyroid gland, we found the fistula usually directly along the normal tissue space around the cricothyroid joint. Accordingly, we removed it together with the surrounding scar tissue without dissecting the recurrent laryngeal nerve.

For patients who also had combined ipsilateral thyroiditis or whose scars were obvious around the thyroid gland, we removed the entire fistula and the surrounding scar tissue and the involved upper lobe of the thyroid. When scar hyperplasia and unclear anatomy are caused by inflammation, the fine anatomy around the cricothyroid joint must be considered when removing the thyroid. It is important to properly protect the recurrent laryngeal nerve, superior laryngeal nerve, and parathyroid glands. To locate the recurrent laryngeal nerve, we look for it within 1 cm of the cricothyroid joint (such as the deep surface of the hypopharyngeal constrictor, or the inferior thyroid artery acting as a marker). Alternatively, we could also search for it along the tracheoesophageal sulcus. After locating the position of the (D)



FIGURE 2 Internal pyriform sinus fistulectomy. (\rightarrow : *fistula*). (A) Making a circular incision around the mouth of the fistula by the CO₂ laser; (B) Separating along the tissue space outside the fistula and cut off it; (C) Cutting and vaporizing the residual fistula tissue by the CO₂ laser; (D) Placing an absorbable gelatin sponge with staining agent in the bottom of the surgical wound; (E) Sewing by the 7-0/8-0 absorbable line.

(E)

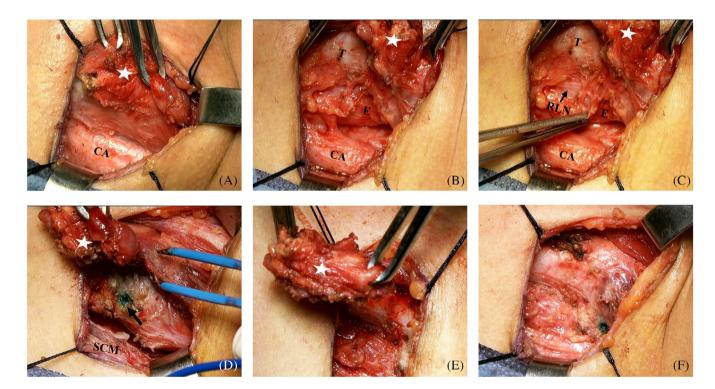


FIGURE 3 External pyriform sinus fistulectomy. (T: trachea; E: esophagus; RLN: recurrent laryngeal nerve; SCM: Sternocleidomastoid muscle; CA: carotid artery; \bigstar : the fistula, involved thyroid lobes and corresponding scar tissue). (A and B) Dissecting the fistula along the scar and normal tissue gap; (C) Dissection the RLN and protect it; (D) Tracing the fistula along its longitudinal axis to the "blue" mark; (E) The removed *fistula*, *involved thyroid lobes*, and *corresponding scar tissue*; (F) Wound condition after removal of fistula.

recurrent laryngeal nerve, we dissected it and protected it (Figure 3C). We were careful to avoid the recurrent laryngeal nerve and its nourishing blood vessels, which are easily damaged. It was important to maintain the integrity of its nourishing blood vessels.

After complete removal of the fistula and its affected tissues, we repeated confirmation that there was no active bleeding and then applied a purse-string suture to seal the wound. Figure 3E,F showed the specimen and the wound condition after the removal of the fistula. Finally, we placed a silicone drainage tube in the surgical field, which was connected to a negative pressure drainage bottle.

2.9 | Postoperative management

All patients received intravenous prophylactic antibiotics at induction and a nasogastric liquid diet six h after surgery. Three days after the operation, budesonide (1 mg, twice a day) was inhaled to exert anti-spasmodic, anti-inflammatory, and anti-asthmatic effects, making it easier for the patient to expectorate sputum, keep the airway open, and decompress. On the eighth day after the operation, patients underwent esophagography. If the results showed no abnormality, the nasogastric tube was removed. The drainage tube was removed when the drainage volume was less than 15 ml in 24 h. A retrospective analysis of surgical time, blood loss, gastric tube residence time, complications, length of the surgical incision, and length of hospital stay was performed. All patients Laryngoscope Investigative Otolaryngology 2149

were followed up and performed video electronic laryngoscopy every three months after surgery.

2.10 | Statistics

The statistical analysis was conducted with the SPSS 22.0 software (IBM Corp, Armonk, NK). Categorical variables were presented as frequencies and proportions. A Chi-square test or Fisher exact test was performed for comparison between the two groups of categorical variables. The student's *t*-test was performed for comparison of measurement data. Data were expressed as mean \pm standard deviation (SD). A two-tailed *p*-value <0.05 was considered statistically significant.

3 | RESULTS

3.1 | Overall outcome

The surgery was successfully completed in 36 patients. One patient was excluded because of the inability to fully expose the pyriformis fossa under the suspension laryngoscopy. The clinical characteristics of 36 patients are shown in Table 1. Of the 36 patients, there were 22 females and 14 males with a mean age of 15 years (range 2–58 years). In all, 32 patients had a history of multiple incisions and drainage, 16 patients had a history of surgical resections, and two

Preoperative variable	$\begin{array}{l} MCWCS^{a} \text{ group} \\ (n=36) \end{array}$	CV ^b group (n = 14)	p value
Demographic			
Female [n (%)]	22 (61.1%)	6 (42.9%)	0.343
Age [mean (range)]	15 (2y-58y)	20 (2y-47y)	0.280
Site of presentation (n [%])			0.326
Left	28 (77.8%)	13 (92.9%)	
Right	3 (8.3%)	1 (7.1%)	
Bilateral	5 (13.9%)	0 (0%)	
Initial presentation (n [%])			
Recurrent low-neck abscess	36 (100%)	14 (100%)	0.071
Acute suppurative thyroiditis	26 (72.2%)	6 (42.9%)	0.099
Neck mass	11 (30.6%)	6 (42.9%)	0.511
History of treatment (n [%])			
Incision and drainage	32 (88.9%)	10 (71.4%)	0.197
Open neck surgery	16 (44.4%)	5 (35.7%)	0.752
Endoscopic cauterizing surgery	2 (5.6%)	0 (0%)	1.000
None	2 (5.6%)	4 (28.6%)	0.044
Positive rate of investigations [n (%)]			
CT enhanced scan	33 (91.7%)	12 (85.7%)	0.611
Laryngoscopy	34 (94.4%)	9 (64.3%)	0.014*

^aMCWCS: CO_2 laser-assisted microsurgery combined with minimally invasive cervical surgery. ^bCV: conventional surgery.

TABLE 1 Demographic and clinical characteristics of patients with different surgical methods

²¹⁵⁰ Investigative Otolaryngology-

$MCWCS^{a}$ group (n = 36)	${\sf CV}^{\sf b}$ group (n = 14)	p value
4.3 ± 0.6	5.5 ± 0.8	<0.0001*
103.2 ± 11.4 ^c	196.8 ± 42.8	<0.0001*
8.1 ± 4.8	27.1 ± 10.7	< 0.0001*
3.2 ± 1.4	5.6 ± 3.8	0.039*
0 (0%)	2 (14.3%)	0.074
0 (0%)	0 (0%)	NA
0 (0%)	0 (0%)	NA
0 (0%)	0 (0%)	NA
2 (5.6%)	1 (7.1%)	1.000
1 (2.8%)	0 (0%)	1.000
7.3 ± 2.6	8.1 ± 3.7	0.399
0 (0%)	4 (28.6%)	0.004*
	4.3 ± 0.6 103.2 ± 11.4^{c} 8.1 ± 4.8 3.2 ± 1.4 $0 (0\%)$ $0 (0\%)$ $0 (0\%)$ $0 (0\%)$ $2 (5.6\%)$ $1 (2.8\%)$ 7.3 ± 2.6	4.3 ± 0.6 5.5 ± 0.8 103.2 ± 11.4^{c} 196.8 ± 42.8 8.1 ± 4.8 27.1 ± 10.7 3.2 ± 1.4 5.6 ± 3.8 0 (0%) $2 (14.3\%)$ 0 (0%) $0 (0\%)$ 0 (0%) $0 (0\%)$ 0 (0%) $0 (0\%)$ 0 (0%) $0 (0\%)$ 1 (2.8%) $0 (0\%)$ 7.3 \pm 2.6 8.1 ± 3.7

LI FT AL.

 a MCWCS: CO $_{2}$ laser-assisted microsurgery combined with minimally invasive cervical surgery.

^bCV: conventional surgery.

^cValues indicate excluding the interval time.

^dValues are expressed as mean ± SD.

patients had a history of cauterization of the internal fistula. In addition, 26 patients presented with thyroid abscesses.

3.2 | Diagnostic investigation

All patients used video electronic laryngoscopy to examine the internal fistula of the pyriform fossa. Finding the internal fistula through laryngoscopy is the premise for our operation to be carried out smoothly. It is worth mentioning that the positive rate was significantly increased up to 94.4% (34/36) after the method of electronic laryngoscopy was revised. In addition, thin-slice CT combined with ultrasonography to assess the shape of the fistula and its relationship to the thyroid is critical for the formulation of a surgical approach. The sensitivity of the fistula on enhanced thin-slice CT scans after oral administration of a contrast agent was 91.7% (33/36), and the sensitivity of thyroid abscess detection by ultrasound thyroid scan was nearly 100% (36/36). These all help the surgeon to perform the surgery successfully.

3.3 | Operative findings

Generally, we found that the fistulas penetrated the hypopharyngeal constrictor along the outer edge of the lower corner of the thyroid cartilage. Interestingly, the fistulas of two patients bypassed the front of the cricothyroid joint before piercing through the hypopharyngeal constrictor. In addition, the fistulas of 26 patients passed through the upper thyroid gland, which resulted in a local abscess of the thyroid gland, and those of 10 patients terminated on the surface of the thyroid lobe. Correspondingly, we performed thyroid lobectomy on these 26 patients with thyroid abscesses. In all cases, the

mean surgical incision length was 4.3 cm. The mean surgical time was 103.2 min, including step 1, which was 34.7 min and step 2 was 68.5 min. The mean blood loss was 8.1 ml. The comparison of surgical outcomes between the MCWCS (CO₂ laser-assisted microsurgery combined with minimally invasive cervical surgery) and CV (conventional surgery) groups are shown in Table 2. Compared with traditional surgery, not only the surgical incision (4.3 cm vs. 5.5 cm, p<0.0001), blood loss (8.1 ml vs. 27.1 ml, p<0.0001) and operation time (103.2 min vs. 196.8 min, p<0.0001) in MCWCS group have been significantly reduced.

3.4 | Postoperative situation

The comparison of postoperative outcomes between the MCWCS and CV groups is shown in Table 2. All patients in the MCWCS and CV group performed the same preoperative and postoperative management. In all cases, the mean gastric tube residence time was seven days. The MCWCS group had a significantly shorter drainage tube residence time (3.2 days vs. 5.6 days, p = 0.002) than the CV group. In the MCWCS group, only one case of anterior neck hematoma occurred after the operation and was treated successfully with no sequelae. Moreover, there were no patients with pharyngeal fistula, permanent recurrent laryngeal nerve paralysis, or hypothyroidism.

3.5 | Follow-up

The mean follow-up duration after excision of the lesion was 34.1 months (median 33 months, range 15–56 months) in the MCWCS group, whereas in the CV group was 57.9 months

(median 60 months, range 39–60 months). There was no recurrence of the fistula in any of the patients in the MCWCS group. In the CV group, a total of four patients relapsed and the mean duration of recurrence was 8.8 months. Compared to the CV group who received conventional surgery, the recurrence rate in the MCWCS group is significantly reduced.

4 | DISCUSSION

Pyriform sinus fistula is a congenital branchial lesion.²⁶⁻²⁸ It has a complicated shape and often originates from the tip or base of the pyriform fossa.^{29,30} Importantly, it is closely related to important structures such as the inferior thyroid cartilage, recurrent laryngeal nerve, and thyroid.³¹⁻³³ Generally, pyriform sinus fistula presents as repeated neck abscesses and acute purulent thyroiditis, which usually causes missed diagnoses.³⁴⁻³⁶ Electronic laryngoscopy is the most intuitive way to observe the pyriform fossa. However, for traditional endoscopy, it is difficult to expose the whole bottom of the pyriform fossa. Moreover, the fistula is usually narrow and located deep in a hidden position, which can easily lead to a missed diagnosis.^{37,38} Accordingly, some scholars have proposed the modified Killian's method used for endoscopic examination, which could fully expose the pyriform fossa area and help with the diagnosis of pyriform sinus fistula.^{39,40} However, this method not only has higher requirements for the patient's body position coordination and movement coordination but also requires higher technical proficiency of the operator, which leads to certain limitations in the implementation of this method. With our understanding of pyriform sinus fistula deepening, we found a new method that could be more easily applied. The patient only needs to be in the supine position and to perform a balloon-blowing action, which is easy to implement even in children. If needed, the operator can lift the larynx to assist in the exposure. Generally, the whole pyriform fossa can be revealed through this method. After applying this method, the positive rate of pyriform sinus fistula diagnosis significantly increased to 95%. Furthermore, enhanced thin-slice CT enhanced scans after oral administration of a contrast agent and ultrasound were used to assist in diagnosing pyriform sinus fistulas and judging the stroke of the fistulas, which could help us formulate surgical strategies.

Since the 1990 s, some scholars have proposed the method of internal fistula cauterization to treat patients with pyriform sinus fistula.⁴¹⁻⁴⁵ However, this method did not remove the fistula epithelial tissue actually and left a closed cavity, where the residual epithelium continued to secrete, fall off, and accumulate. These factors caused the disease to relapse when the patients, especially patients with thyroiditis, have low resistance or when they acquire a secondary infection. Therefore, complete removal of the fistula is the most effective way to reduce its recurrence. Traditional open surgery through the neck can achieve resection of the fistula.^{1,46} However, it is difficult to identify the fistula structure during surgery, which makes it difficult to resect completely.⁵ In addition, some scholars advocate the use of some materials (such as zebra guidewire) to identify the

location of the fistula during the operation,⁴⁷ but its use is limited, and the guidewire needs to be removed after the fistula is removed, which increases the complexity of the surgical procedure. Based on this, we advocate a new surgical procedure to treat pyriform sinus fistula, especially for complex fistulas. First, we used a CO₂ laser to resect the internal fistula. Because the CO₂ laser had high accuracy and low thermal damage, we could completely remove the internal fistula along the interstitial space. Moreover, relying on the direct visibility with the endoscope, we could place a gelatin sponge stained with methylene blue on the bottom of the wound, which we used to locate the position of the fistula for subsequent surgery. Because the dye did not diffuse, the sponge helped us accurately locate the fistula, providing the possibility for a subsequent minimally invasive cervical surgery to completely resect the fistula. In addition, because the sponge was absorbable, it was not necessary to remove the marker. After clarifying the location of the fistula, we could achieve a complete resection of the fistula by using a surgical method similar to thyroid lobectomy. If we found that the fistula was limited to the cricothyroid joint and did not pass through the thyroid gland through preoperative evaluation, we only needed to perform the first step to achieve the fistulectomy.

In our study, most patients were referred to our hospital due to recurrence. Among them, 32 patients recurred after repeated incision and drainage, 16 patients recurred after resection of the fistula through the neck approach, two patients recurred after cauterizing the internal fistula via endoscopy, and some of them experienced more than two or three of the above treatments. Through this surgical approach, we could not only cure recurrent pyriform sinus fistula but also remove the previous scar, satisfying the patient's requirements for the aesthetic appearance of the neck. Moreover, compared with traditional surgery, not only the surgical incision, blood loss and operation time with this surgery have been significantly reduced. In addition, none of the 36 patients in this study had complications such as pharyngeal fistula, permanent recurrent laryngeal nerve paralysis, or hypothyroidism. The mean follow-up duration after excision of the lesion was 34.1 months (median 33 months, range 15-56 months) and no patients who performed this surgery have relapsed.

This surgical technique is simple and can be self-taught. If the conditions of CO₂ laser equipment are not available, a long electrocautery or electrocoagulation knife can be used to cauterize the internal fistula instead. Although a little fistula tissue will remain, the recurrence of pyriform fossa fistula can be greatly reduced. When excising the pyriform fistula through the external cervical approach, care should be taken to properly protect the recurrent laryngeal nerve, avoid traction injury as much as possible, and try to maintain the integrity of its nourishing blood vessels. However, there were some limitations worthy to be of consideration in this study. Compared with the CV group, the shorter length of follow-up in the MCWCS group may account for the decrease in recurrence. Moreover, finding the internal fistula under the suspension laryngoscope and endoscope is the key to this operation. For patients who cannot be fully exposed under suspension laryngoscopy, the surgery is hard to perform.

5 | CONCLUSION

Our experience showed that this surgical technique could be used to completely remove the fistula, especially for complex pyriform fistulas. This operation was easy to perform and took less time than the conventional strategies. Through this surgery, the patient had less trauma, the neck had fewer scars, and the recurrence rate was significantly reduced. Therefore, we recommend using this surgical technique to treat patients with complicated pyriform fistulas.

ACKNOWLEDGMENTS

The authors would like to thank AJE for the English language editing.

FUNDING INFORMATION

This work was supported by the National Key R&D Program of China (No. 2020YFC1316903), the 5010 Clinical Research Program of Sun Yat-sen University (No. 2017004), and the General Program of the National Natural Science Foundation of China (No. 81972528).

CONFLICT OF INTEREST

All authors declare that there is no conflict of interest associated with this study.

ORCID

Wenbin Lei 🕩 https://orcid.org/0000-0002-9720-1997

REFERENCES

- James A, Stewart C, Warrick P, Tzifa C, Forte V. Branchial sinus of the piriform fossa: reappraisal of third and fourth branchial anomalies. *Laryngoscope*. 2007;117:1920-1924.
- Nicoucar K, Giger R, Pope HG Jr, Jaecklin T, Dulguerov P. Management of congenital fourth branchial arch anomalies: a review and analysis of published cases. J Pediatr Surg. 2009;44:1432-1439.
- Kakogawa J, Nako T, Igarashi S, Nakamura S, Tanaka M. Prenatal diagnosis of pyriform sinus fistula: case report and literature review. *Clin Case Rep.* 2015;3:106-109.
- Cai Q, Pan Y, Yaodong X, et al. Resection of recurrent branchial cleft deformity using selective neck dissection technique. *Int J Pediatr Otorhinolaryngol.* 2014;78(7):1071-1073.
- Derks LS, Veenstra HJ, Oomen KP, Speleman L, Stegeman I. Surgery versus endoscopic cauterization in patients with third or fourth branchial pouch sinuses: a systematic review. *Laryngoscope*. 2016;126: 212-217.
- Chen T, Chen J, Sheng Q, et al. Pyriform sinus fistula in children: a comparison of endoscopic-assisted surgery and endoscopic radiofrequency ablation. J Pediatr Surg. 2021;56:800-804.
- Chen W, Chen J, Chen F, et al. Endoscopic coblation treatment for congenital pyriform sinus fistula in children. *Medicine (Baltimore)*. 2021;100:e25942.
- Watson GJ, Nichani JR, Rothera MP, Bruce IA. Case series: endoscopic management of fourth branchial arch anomalies. *Int J Pediatr Otorhinolaryngol.* 2013;77:766-769.
- Yanagisawa S, Oshio T, Kato M, Kano M, Tsuji Y, Morikawa Y. Endoscopic chemocauterization for pyriform sinus fistula in children. *Pediatr Int*. 2017;59:807-811.
- Cuestas G, Doormann F, Rodriguez V, Bellia Munzon P, Bellia MG. Endoscopic electrocauterization for congenital pyriform sinus fistula treatment in paediatrics. *Case Ser Acta Otorrinolaringol Espanol.* 2017; 68:220-225.

- 11. Sheng Q, Lv Z, Xu W, Liu J. Reoperation for pyriform sinus fistula in pediatric patients. *Front Pediatr.* 2020;8:116.
- 12. Wang WC, Shen HN. Thyroid abscess associated with pyriform sinus fistula. *Mayo Clin Proc.* 2021;96:2854-2855.
- Hao Z, Hou Y, Li X, et al. Misdiagnosis of asymptomatic intrathyroidal pyriform sinus fistula: a case report. J Int Med Res. 2021;49: 3000605211031430.
- 14. Kondo T. Acute suppurative thyroiditis secondary to pyriform sinus fistula. *Lancet Infect Dis*. 2019;19:447.
- 15. Pan J, Zou Y, Li L, Yang TY, Yang JL, Hu C. Clinical and imaging differences between neonates and children with pyriform sinus fistula: which is preferred for diagnosis, computed tomography, or barium esophagography? *J Pediatr Surg.* 2017;52:1878-1881.
- Zhao L, Chen L, Li W, Ni K, Chen W, Li X. Retrospective review of 70 cases of pyriform sinus fistula. *Int J Pediatr Otorhinolaryngol.* 2021; 150:110904.
- 17. Han Z, Tai J, Gao J, et al. MRI in children with pyriform sinus fistula. *J Magn Reson Imaging*. 2021;53:85-95.
- Nicoucar K, Giger R, Jaecklin T, Pope HG Jr, Dulguerov P. Management of congenital third branchial arch anomalies: a systematic review. Otolaryngology–Head Neck Surg Off J Am Acad Otolaryngol-Head Neck Surg. 2010;52:1989-1993.
- Chen T, Chen J, Sheng Q, Zhu L, Lv Z. Pyriform sinus fistula in the fetus and neonate: a systematic review of published cases. *Front Pediatr.* 2020;8:502.
- Zhu H, Xiao X, Zheng S, Shen C. Diagnosis and management of pyriform sinus cyst in neonates: 16-year experience at a single center. *J Pediatr Surg.* 2017;52:1989-1993.
- 21. Sheng Q, Lv Z, Xiao X, et al. Diagnosis and management of pyriform sinus fistula: experience in 48 cases. *J Pediatr Surg.* 2014;49:455-459.
- 22. Sai Prasad TR, Chong CL, Mani A, et al. Acute suppurative thyroiditis in children secondary to pyriform sinus fistula. *Pediatr Surg Int.* 2007; 23:779-783.
- Madana J, Yolmo D, Gopalakrishnan S, Saxena SK. Cervical infection secondary to pyriform sinus fistula of branchial origin. *Congenit Anom.* 2009;49:276-278.
- 24. Mou JW, Chan KW, Wong YS, Houben CH, Tam YH, Lee KH. Recurrent deep neck abscess and piriform sinus tract: a 15-year review on the diagnosis and management. *J Pediatr Surg.* 2014;49:1264-1267.
- 25. Zhang P, Tian X. Recurrent neck lesions secondary to pyriform sinus fistula. *Eur Arch Otorhinolaryngol.* 2016;273:735-739.
- Thomas B, Shroff M, Forte V, Blaser S, James A. Revisiting imaging features and the embryologic basis of third and fourth branchial anomalies. *AJNR Am J Neuroradiol.* 2010;31:755-760.
- 27. Liberman M, Kay S, Emil S, et al. Ten years of experience with third and fourth branchial remnants. *J Pediatr Surg.* 2002;37:685-690.
- Lin CY, Hsiao TY, Hsu WC. Pathognomonic ultrasonic images of congenital pyriform sinus fistula in children. *Clin Otolaryngol.* 2021;46:110-115.
- Li Y, Lyu K, Wen Y, et al. Third or fourth branchial pouch sinus lesions: a case series and management algorithm. J Otolaryngol-Head Neck Surg. 2019;48:61.
- Chen T, Ge G, Chen J, et al. Pyriform sinus fistula in children: preferred imaging modality and risk factors for diagnostic delay. *Front Pediatr.* 2020;8:575812.
- Josephson GD, Black K. A review over the past 15 years of the management of the internal piriform apex sinus tract of a branchial pouch anomaly and case description. Ann Otol Rhinol Laryngol. 2015;124:947-952.
- Zhu T, Huang Y. Contrast-enhanced ultrasound diagnosed a pyriform sinus fistula that failed to be diagnosed by a barium swallow examination: case report and review of choices for the imaging examination. *J Ultrasound Med.* 2020;39:1669-1673.
- Wang S, Mei L, Li Y, et al. Application of gastroscopy in the diagnosis of congenital pyriform sinus fistula in children. Front Pediatr. 2021;8:541249.
- Mali VP, Prabhakaran K. Recurrent acute thyroid swellings because of pyriform sinus fistula. J Pediatr Surg. 2008;43:27-30.

Laryngoscope Investigative Otolaryngology 2153

- Lafontaine N, Learoyd D, Farrel S, Wong R. Suppurative thyroiditis: systematic review and clinical guidance. *Clin Endocrinol (Oxf)*. 2021; 95:253-264.
- Yang H, Li D, Ye X, et al. Aspiration with or without lavage in the treatment of acute suppurative thyroiditis secondary to pyriform sinus fistula. Arch Endocrinol Metab. 2020;64:128-137.
- 37. Li L, Zhao DJ, Yao TY, et al. Imaging findings in neonates with congenital pyriform sinus fistula: a retrospective study of 45 cases. *Front Pediatr.* 2021;9:721128.
- Sheng Q, Lv Z, Xu W, Liu J. Differences in the diagnosis and management of pyriform sinus fistula between newborns and children. *Sci Rep.* 2019;9:18497.
- Kano M, Murono S, Yamamoto T, Yoshizaki T. Preoperative identification of the internal opening with the modified Killian's method in a case of pyriform sinus fistula. *Am J Otolaryngol.* 2016;37:38-40.
- Huang SL, Chen LS, Zhang B, et al. Value of modified Killian's method in diagnosis of congenital pyriform sinus fistula. *Zhonghua Er Bi Yan Hou Tou Jing Wai Ke Za Zhi*. 2017;52:744-748.
- Chen LL, Liang XL, et al. Preliminary experiences of endoscopic CO₂ laser cauterization for treatment of congenital pyriform sinus fistula. Zhonghua Er Bi Yan Hou Tou Jing Wai Ke Za Zhi. 2014;49: 582-585.
- 42. Pereira KD, Smith SL. Endoscopic chemical cautery of piriform sinus tracts: a safe new technique. *Int J Pediatr Otorhinolaryngol.* 2008;72:185-188.
- Masuoka H, Miyauchi A, Sasaki T, Sano T, Miya A. Outcomes of endoscopic chemo- and laser-cauterizations and open fistulectomy for pyriform sinus fistula. J Otolaryngol Head Neck Surg. 2021;50:49.

- 44. Huang YC, Peng SS, Hsu WC. KTP laser assisted endoscopic tissue fibrin glue biocauterization for congenital pyriform sinus fistula in children. *Int J Pediatr Otorhinolaryngol.* 2016;85:115-119.
- 45. Di Nardo G, Valentini V, Angeletti D, et al. Recurrent pyriform sinus fistula successfully treated by endoscopic Glubran 2 sealing: a rare case and literature review. SAGE Open Med Case Rep. 2016;4: 2050313X16672151.
- Nicollas R, Ducroz V, Garabédian EN, Triglia JM. Fourth branchial pouch anomalies: a study of six cases and review of the literature. *Int J Pediatr Otorhinolaryngol.* 1998;44:5-10.
- Ding XQ, Zhu X, Li L, Xu F, Huang ZC. Resection of recurrent third branchial cleft fistulas assisted by flexible pharyngotomy. World J Clin Cases. 2019;7:3957-3963.

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

How to cite this article: Li Y, Yue H, Lei G, et al. A safe and effective surgical method for complex pyriform sinus fistula. *Laryngoscope Investigative Otolaryngology*. 2022;7(6): 2145-2153. doi:10.1002/lio2.971