Endoscopic diagnosis and treatment of a pyriform sinus-cutaneous fistula in a non-pediatric patient: thinking outside the box



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Background and Aims: Pyriform sinus-cutaneous fistula is often observed in pediatric patients and is very rare in adults. Diagnosing this condition in adults is challenging, and contrast-enhanced CT misdiagnosis is not uncommon. Neck abscess formation secondary to this fistula is treated with surgical incision and drainage, but recurrence can occur due to the fistula. Although surgery is the most effective treatment modality, less-invasive therapies such as electrocauterization or chemocauterization may be indicated. However, treatment with these cauterization techniques has a high treatment failure rate.

Methods: This is the first case report of diagnosing, by EGD, a pyriform sinus fistula in an adult treated with autologous abdominal fat transplantation associated with adjunctive conventional endoscopic therapies.

Results: A 35-year-old woman with a history of 2 cervical abscesses was admitted due to a recurrent abscess with unknown etiology. She underwent surgical drainage with intraoperative EGD under fluoroscopic assistance, and a pyriform sinus-cutaneous fistula was diagnosed. Subsequently, fistula treatment with unprocessed autologous adipose abdominal tissue collected by lipoaspiration associated with argon plasma coagulation and endoscopic vacuum therapy were successfully performed. Immediately after the procedure, the patient no longer experienced fluid discharge through the skin. The patient had no recurrence within 10 months of follow-up.

Conclusions: Autologous abdominal fat transplantation may be an effective minimally invasive therapy for pyriform sinus-cutaneous fistula and has the potential to become an alternative therapy for GI fistulas. We encourage future studies to exploit the role of this therapy for GI fistulas. (iGIE 2024;3:48-52.)

Pyriform sinus-cutaneous fistula (PSCF) is often observed in pediatric patients and is rare in adults. The diagnosis of this condition in adults is challenging, and contrast-enhanced CT misdiagnosis is not uncommon.¹

Neck abscess formation secondary to this fistula is treated with surgical incision and drainage, but recurrence can occur due to the fistula.² Surgery is the most effective treatment modality, but less-invasive therapies such as electrocauterization or chemocauterization can be indicated as a primary or rescue therapy.³

This article describes a promising minimally invasive technique for managing complex fistulas.

METHODS

To the best of our knowledge, this is the first case report of a PSCF in an adult diagnosed by EGD under fluoroscopic assistance and treated with autologous abdominal fat transplantation. Argon plasma coagulation (APC) and endoscopic vacuum therapy (EVT) were used as adjunctive therapies. The first therapy aims to loosen granulation tissue before applying the abdominal fat, and the last to avoid contact of the saliva with the treated area by fluid aspiration.^{4,5}

Because the lipoaspirate tissue is an abundant source of adipocytic-derived stem cells, autologous fat transfer is considered an ideal soft tissue filler. These cells can integrate into the host tissue and secrete a relevant orchestrated portion of cytokines and growth factors, including vascular endothelial growth factor, hepatocyte growth factor, insulin-like growth factor, platelet-derived growth factor, and transforming growth factor β .^{6,7}

RESULTS

A 35-year-old woman with a history of 2 cervical abscesses was admitted due to a recurrent abscess with unknown etiology after imaging examinations. Physical examination



Figure 1. Small fistulous orifice in the pyriform sinus.

identified an area of tenderness with erythema, swelling, and warmth in the anterior neck with purulent discharge.

She underwent surgical drainage with intraoperative EGD under fluoroscopic assistance and a small orifice (Fig. 1) in the pyriform sinus. A short, thin, and tortuous tract to a cutaneous orifice in the neck was diagnosed (Fig. 2).

After lipoaspiration (Fig. 3), fistula treatment was then performed. A .035-inch guidewire was placed through the fistula; the APC probe was attached to the guidewire from the skin and then pulled through the mouth. It was then pulled back through the skin, applying electrocauterization in the fistula tract and its margins (Fig. 4). After de-epithelization, unprocessed autologous adipose abdominal tissue collected by lipoaspiration (Fig. 5) was injected through the pyriform sinus orifice and its margins using a 21-gauge injection needle under endoscopic visualization, followed by filling the fistula tract with fat (Fig. 6). Finally, an EVT system manufactured in the gastric portion of a triple lumen tube was placed, allowing both nutrition and aspiration with 1 tube through the nostril (Fig. 7).^{5,8} Although achieving negative pressure is required for proper EVT function, EVT was primarily used for fluid aspiration in this case. Immediately after the procedure, the patient no longer experienced fluid discharge through the skin. The triple lumen tube was removed after 1 week. Three-month follow-up EGD under fluoroscopic assistance and orotracheal intubation confirmed successful closure (Fig. 8). The patient did not present recurrence during 10 months of follow-up (Fig. 9).

DISCUSSION

Surgical drainage and antibiotic therapy remain the criterion standard therapy for managing PSCF-associated abscesses. However, less-invasive approaches are needed,



Figure 2. Pyriform sinus-cutaneous fistula with a short, thin, and tortuous tract.



Figure 3. Lipoaspiration.

mainly when treatment failure occurs. Although hypopharynx fistulas are not usually treated by GI endoscopists,⁹ those with prior experience in transmural GI defect closures may have the ability to treat this condition.

Administration of unprocessed adipose tissue has numerous advantages compared with lipoaspirate by enzymatic methods. These advantages include minimal manipulation and lower cost; in addition, it is less time-consuming and can be performed intraoperatively.¹⁰



Figure 4. Fistulous tract de-epithelization before autologous abdominal fat transplantation. **A**, Placement of a .035-inch guidewire through the fistula tract assisted by fluoroscopic and endoscopic images. **B**, Adapted argon plasma coagulation probe delivery system using a .035-inch guidewire and a surgical drape. **C**, Post-electrocauterization of the fistulous tract and its margins.

Figure 5. Unprocessed autologous adipose abdominal tissue collected by lipoaspiration. **A**, Unprocessed adipose abdominal tissue immediately after acquisition by lipoaspiration. **B**, Unprocessed adipose tissue introduced in a 20 mL syringe. **C**, Adipose tissue transfer to a 3 mL syringe. **D**, Unprocessed autologous adipose abdominal tissue in a 3 mL syringe ready for use.

Figure 6. Unprocessed autologous adipose abdominal tissue for the treatment of a pyriform sinus-cutaneous fistula. A, Endoscopic image after the procedure. B, Cutaneous orifice occluded by the unprocessed adipose abdominal tissue.

Figure 7. Endoscopic vacuum therapy as an adjunctive therapy for fistula closure. **A**, Endoscopic vacuum therapy system manufactured with a triple lumen tube and a polyurethane open-pore sponge connected to the gastric (aspiration) portion of the tube. **B**, Modified endoscopic vacuum system placement.

Figure 8. Imaging confirms successful fistula closure and the importance of orotracheal intubation. **A**, Endoscopic image shows a complete closure of the pyriform sinus orifice with a white scar. **B**, Water-soluble contrast injection through the gastroscope working channel confirming no extravasation. **C**, Orotracheal tube balloon (cuff) preventing aspiration, showing the importance of general anesthesia during this procedure.

Figure 9. Cutaneous orifice healing process. A, Image immediately after the procedure. B, Follow-up image at 15 days. C, Follow-up image at 45 days. D, Follow-up image at 120 days.

In conclusion, autologous abdominal fat transplantation is feasible and seems to be safe and effective for PSCF closure. This strategy may also be effectively applied to GI fistulas and has the potential to become an alternative therapy for this challenging condition. We encourage future studies to explore the role of this therapy for GI fistulas.

DISCLOSURE

All authors disclosed no financial relationships.

Abbreviations: APC, argon plasma coagulation; EVT, endoscopic vacuum therapy; PSCF, pyriform sinus-cutaneous fistula.

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