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Case Report
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Submucosal Tunneling Endoscopic Septum Division for Congenital Pseudo-Pylorus: A New Technology in a Rare Disease (With Video)

Xinyang Liu[1], Ying Fang[2], Hong-Bin Yang[2], Xiao-Xia Ren[2], Ya-Nan Han[2], Quan-Lin Li[1] and Ping-Hong Zhou[1]**

[1]Department of Endoscopy, Fudan University, China

[2]Department of Gastroenterology, The Children's Hospital of Xi'an City, China

Abstract

Background: Pseudo-pylorus is a congenital or an acquired septum (secondary to ulcer scaring) in the antral area that might simulate the pylorus. It could easily cause obstruction thus requires intervention. We report the use of Submucosal Tunneling Endoscopic Septum Division (STESD) on a pediatric case of congenital pseudo-pylorus.

Methods: This video illustrates the STESD procedure in an 8-year-old Chinese boy. With a congenital pseudo-pyloric stricture that occurred proximal to the original pylorus, he was suffering from recurrent vomiting, epigastric pain and growth retardation. A gastroscopy demonstrated pseudo-pyloric stricture with severe reflux esophagitis. STESD was performed according to the standard protocol (step 1: mucosal incision; step 2: submucosal tunneling; step 3: septum division; step 4: mucosal closure.) with appropriate preoperative and postoperative care.

Results: The operation time was 35minutes.Intraoperative pneumoperitoneum was relieved by paracentesis. The pseudo pylorus disappeared and symptoms were completely relieved after the procedure. The patient was discharged uneventfully at postoperative day 10. In four-month follow-up, we observed complete symptom resolution, significant improvement of nutritional status and no complications.

Conclusion: We suggest STESD as a safe and efficient technology in treating congenital pseudo-pylorus. Validation in other benign strictures of the gastrointestinal tract is warranted.

Keywords: STESD; Pseudo-pylorus; Congenital; Endoscopic treatment

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Correspondence:
Ping-Hong Zhou, Department of Endoscopy, Fudan University, China, Tel: (+86)-21-64041990; Fax: +86-21-64038472; E-mail: zhou.pinghong@zs-hospital.sh.cn

Quan-Lin Li, Department of Endoscopy, Fudan University, China, Tel: (+86)-21-64041990; Fax: +86-21-64038472; E-mail: li.quanlin@zs-hospital.sh.cn

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Introduction

Pseudo-pylorus is a congenital or an acquired septum (secondary to ulcer scaring) in the antral area that might simulate the pylorus [1]. Different from the normal intermittent, ring-like contraction of the pyloric loop, it could easily cause obstruction.

Although in some literature, the term pseudo-pylorus was sometimes confused with "double pylorus" (also known as pyloric diaphragm, antral mucosal band and pyloric septum) [2-3], another rare abnormality usually arising as an acquired complication of chronic peptic ulcer disease and rarely congenital [4-8], the two terms are essentially different.

In double pylorus, the two pylori occur "in parallel", with a gastro duodenal fistula consisting of a short accessory channel between the distal stomach and the duodenal bulb, such that the gastric antrum and the duodenal bulb are connected by two separate openings [4-8]. In this case, it is asymptomatic. However, as the pseudo-pylorus occurs "in series" with the anatomic-pylorus, It is a benign stricture that causes obstructive symptoms and thus requires intervention [1].

Herein, we report the use of a new method recently developed by our institution for the treatment of Zenker's diverticulum----Submucosal Tunneling Endoscopic Septum Division (STESD) [9], for treating a pediatric case of congenital pseudo-pylorus.

Case Presentation

The procedure was performed on an 8-year-old Chinese boy who had been suffering from

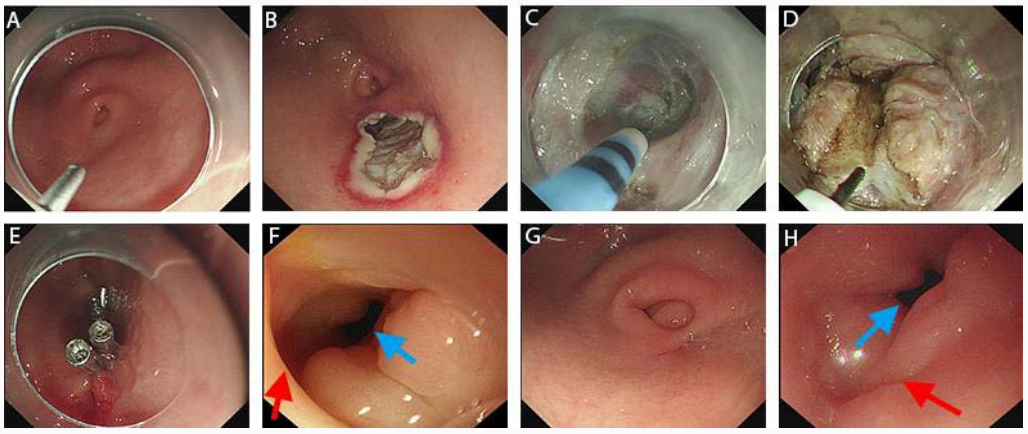

Figure 1: Submucosal Tunneling Endoscopic Septum Division (STESD) for a pediatric case of congenital pseudo-pylorus. (A) Endoscopic view of the pseudo-pylorus. (B) Submucosal injection and mucosal incision 3 cm proximal to the pseudo-pylorus. (C) Creation of the submucosal tunnel and clear exposure of the muscle fibers of the septum inside the tunnel. (D) Septum division under direct endoscopic view. (E) Closure of the mucosal tunnel entry. (F) Endoscopic view of the relaxed pseudo-pylorus (red arrow) and the anatomic pylorus (blue arrow). (G) Follow-up endoscopic view of the relaxed pseudo-pylorus at month 4. (H) Follow-up endoscopic view of the relaxed pseudo-pylorus (red arrow) and the anatomic pylorus (blue arrow) at month 4.

recurrent vomiting, epigastric pain and growth retardation. The past medical history revealed cryptorchism and was otherwise unremarkable. A gastroscopy was performed and demonstrated pyloric stricture with severe reflux esophagitis. A single-channel gastroscope with a diameter of 9.6 mm (GIFQ 260; Olympus Medical Systems Co. Tokyo, Japan) could not pass through the stricture. Interestingly, after passing through the above-mentioned "pyloric" stricture with a 5.8 mm gastroscope (GIF-XP290N; Olympus Medical Systems Co, Tokyo, Japan), another pylorus was observed, implicating that the previous "pylorus" was a congenital pseudo-pylorus that occurred in series with and proximal to the original pylorus. Rapid urease test showed that *H. pylori* was negative. His hemoglobin level was low at 5.82 g/dL, and then corrected by transfusion to 9.0 g/dL preoperatively.

A single-channel gastroscope (GIF-Q260J; Olympus Medical Systems Co, Tokyo, Japan) was used with a hybrid knife (ERBE; Erbe Elektromedizin GmbH, Tubingen, Germany). Carbon dioxide (CO_2) gas was used for insufflation during the procedure with a CO_2 insufflator (UCR; Olympus). A transparent cap (D-201-11802, Olympus) was attached to gastroscope tip. Prophylactic intravenous antibiotics were administered. STESD was performed as follows (Video 1 and Figure 1): Step 1: Mucosal incision. Mucosal entry was made 3 cm proximal to the stricture of the pseudo-pylorus. A 2.0 cm longitudinal mucosal incision was performed at the tunnel entry after injection of saline mixed with indigo carmine and epinephrine.

Step 2: Submucosal tunneling. A submucosal tunnel was created between the mucosal and muscular layers. The tunnel was created from the incision point till 2 cm distal to the stricture to ensure enough working space for myotomy.

Step 3: Septum division. The thickened septal muscle fibers at the stricture were dissected by longitudinal incisions till 2 cm beyond the stricture.

Step 4: Mucosal closure. Mucosal closure of the tunnel entry was made with hemostatic clips after careful hemostasis.

The procedure took 35 mins. Abdominal distention was observed during the procedure, suggesting pneumoperitoneum, and was relieved by paracentesis. No mucosal injury occurred. Intravenous antibiotics were continued for one week postoperatively. Nasal feeding started at postoperative day 7 and the patient was discharged at postoperative day 10. The follow-up gastroscopy 4 months after the operation confirmed the relaxation of the pseudo-pylorus and the patient's symptoms were relieved completely.

Discussion

Previous publications have reported cases with congenital or acquired two pylori "in parallel" [4-8], known as "double pylorus", while pseudo-pylorus with two pylori "in series" has been barely known [1]. In our case, the pseudo-pylorus appeared "in series" with the anatomic pylorus, causing recurrent vomiting and subsequent severe malnutrition due to the stricture. Therefore, surgical intervention was necessary. Pseudo-pylorus usually forms secondary to a peptic ulcer scar. However, this boy had no history of peptic ulcer, and gastroscopy also detected no *H. pylori* infection or peptic ulcer scar, suggesting the congenital origination of this condition. The pathogenesis of this condition is unknown and we propose the imbalance between contraction and relaxation of the circular muscular fibers might be one explanation.

STESD is a novel technique recently developed by our institution for the treatment of Zenker's diverticulum [9]. It was inspired by the peroral endoscopic myotomy (POEM) technique [10]. Theoretically, it could release the stricture of the gastrointestinal tract with maintenance of mucosal integrity when selectively dividing the muscular septum, which in this case equals the thickened circular muscular fibers at the pseudo-pylorus. Therefore, it advantages in possibly reducing the risk of postoperative perforation and secondary infection. Complete division of thickened muscles and protection of the gastric wall is also facilitated by adequate exposure and enough working space by the submucosal tunnel. In this pediatric case, we believe it is better than balloon dilation, which is not long-lasting and usually requires repeated procedures. In the meantime, it is also less invasive than an open surgery. To our knowledge, this is the first application of STESD in this rare disease. Besides pseudo-pylorus, STESD is also potentially applicable in other conditions of benign stricture of the gastrointestinal tract.

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