CASE REPORT

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Pediatric gastropleural fistula, a complication of sleeve gastrectomy: Case report and brief review

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Abstract

Gastropleural fistula (GPF) is a rare pathological communication between the stomach and pleura. It may complicate sleeve gastrectomy (SG). An endoscopic application of OTSC can be used to manage GPF.

KEYWORDS

endoscopy, gastropleural fistula, over-the-scope clip, sleeve gastrectomy

INTRODUCTION 1

Gastropleural fistula (GPF) can be defined as a pathological communication between the stomach and the pleura that may develop secondary to infection, malignancy, and repair of congenital defects. Recently, it has been reported secondary to sleeve gastrectomy (SG) in the adult population. Here, we describe the first case of pediatric GPF secondary to SG in the literature; the case was complicated by hydropneumothorax and lung abscess. It was managed endoscopically with an over-the-scope clip (OTSC) after failure of surgical repair.

Gastropleural fistula is a communication between the stomach and pleura that may occur in adult patients as a complication of sleeve gastrectomy (SG), a widely accepted technique used in weight-reduction surgery.^{1,2} In addition, it has been reported to be secondary to infection, malignancy, surgery, and trauma. However, GPF is infrequently reported in the pediatric population. In our literature review, we found five reports of pediatric GPF, and none of those cases were related to bariatric weight-reduction surgery (Table 1). We

report the first pediatric GPF secondary to SG, who presented with hydropneumothorax and lung abscess.

2 **CASE DESCRIPTION**

In our case, an 11-year-old boy underwent SG due to morbid obesity (body mass index $[BMI] = 55.6 \text{ kg/m}^2$). Surgery took place in a private hospital outside Saudi Arabia. It was reported as uneventful, and the patient was discharged after 1 week with a nutritional plan and vitamin supplements. Postoperatively, he had no complaints apart from one admission to his local hospital for gastroenteritis and dehydration that was treated with intravenous (IV) fluids and antibiotics.

Five months after the operation, the patient developed fever, followed by productive cough and dyspnea. He was admitted to the local hospital and managed as a case of community-acquired pneumonia based on chest X-ray (CXR) findings of localized left lower lobe opacity. The results of a barium meal performed at that time were reported to be

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	Age	Diagnosis	Presentation	Diagnostic modalities	Attempted endoscopic interventions	Final intervention	Outcome
Prasertsan et al ¹²	8 y	Large B-cell lymphoma	Chylothorax-like pleural effusion	Chest computed tomography (CT) scan	Failed clipping method and surgical glue	Fistula tract closure with intercostal muscle flap	Uneventful recovery
Wuthisuthimethawee et al ¹³	12 y	Pyogenic splenic abscesses, postsplenectomy; <i>Mycobacterium</i> tuberculosis	Persistent empyema, bile- like effusion	Barium meal with water-soluble contrast	N/A	Fistula division with repair of the stomach and left diaphragm	Uneventful recovery
Arun et al ¹⁴	7 y	Chronic granulomatous disease, <i>Candida</i> <i>albicans</i> infection	Loculated empyema	Barium meal and chest CT scan	N/A	Fistula repair with gastrostomy and feeding jejunostomy	Died
Malik et al ¹⁵	Newborn	Congenital diaphragmatic hernia (CDH) with <i>Candida</i> infection	Persisted effusion, greenish color	Endoscopy	Diagnostic, no intervention	Fistula resection and repair	Uneventful recovery
Lakshminarayanan et al ⁸	2.5 y	CDH and fundoplication for gastroesophageal reflux disease (GERD)	Respiratory distress	Laparoscopic visualization	N/A	Surgical closure	Uneventful recovery

TABLE 1 Reported gastropleural fistulas (GPFs) in the pediatric population

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FIGURE 1 This figure shows the improvement in radiological findings. Preendoscopic chest X-ray (A) and CT scan (C) which shows a contrast leak and loculated encysted fluid (Star). And postendoscopic CXR (B) and CT scan with oral contrast (D) which shows a regression of the loculated fluid and no more leak from the fistula

FIGURE 2 Endoscopic application of OVESCO

normal. The patient was not improving, so he was referred to his primary surgeon, who carried out chest computed tomography (CT) with oral contrast; this procedure showed a leak of contrast into the pleural space, indicating the presence of a GPF. Based on these findings, our patient underwent laparoscopic surgery for fistula closure. Intra-operatively, a left lower lobe abscess was found, measuring around 5 cm \times 6 cm. The surgeon converted the procedure to thoracotomy for fistula closure with left lower lobectomy.

After surgery, the patient still had a chronic cough and fluctuating fever, and a follow-up barium meal showed a leak at the site of the fistula. A repeated chest CT scan revealed multiple encysted pockets of fluid within the pleura (Figure 1). These thoracic complications were treated with video-assisted thoracoscopic surgery (VATS) for decortication, and because the patient still had an ongoing leak, oral feeding was not resumed. Instead, a gastrojejunal (GJ) tube was inserted for feeding, and a second tube was introduced through the fistula opening for drainage. The patient was referred to our tertiary center for further management of his persistent GPF. Upon admission, he had a history of fever and chronic dry cough. His initial laboratory workup showed elevated ESR of 120 mm/H. He was managed with empirical broad-spectrum antibiotics (clindamycin and ceftazidime) and kept non per os (NPO) with strict GJ feeding. Four months after the VATS, the patient underwent an endoscopy, which showed a 5 mm fistula opening just below the gastroesophageal junction toward the posterior wall of the gastric fundus, with an edge showing mild erythema. An OTSC was applied to close the fistula using endoscopic suction (Figure 2). The patient had no postprocedural complications and was discharged on GJ feeding and follow-up to ensure that there was no persistent leak.

Four weeks after endoscopy, a barium meal procedure showed no evidence of leak into the pleura, and a follow-up CXR displayed marked improvement in the aeration of the left lung (Figure 1). A repeat CT scan with oral contrast at 8 weeks postendoscopy showed no leak, and it was observed

that the pockets of encysted pleural fluid had regressed (Figure 1). Oral feeding was gradually resumed, and the patient was tolerating this adjustment. His last visit to our hospital was 10 months after his GPF closure. His weight was 54 kg, and he had a BMI of 21 kg/m²; he looked well-nourished and seemed to have recovered completely.

3 | **DISCUSSION**

Gastropleural fistula is a rare, pathological communication between the stomach and the pleural cavity.¹ It was first described by Markowitz and Herter in 1960 as a complication of hiatal hernia rupture.³ Since then, it has been reported secondary to trauma, peptic ulcer disease, and malignancy (lymphoma and gastric tumors), as well as because of the spread of infection from adjacent structures, such as subdiaphragmatic abscess, empyema, or invasive fungal infection, in immunocompromised patients. Recently, with the emergence of bariatric surgeries, GPF has been documented as a complication of such surgeries in adult patients.⁴ One of these surgeries is SG, which has been utilized in pediatric patients to a lesser extent.² Our patient underwent SG; unfortunately, this was complicated by the development of GPF.

Gastropleural fistula exhibits latent presentation, presenting months to years after SG.¹ The symptoms range from chronic cough to recurrent respiratory infections, both of which were seen in our case.¹ Other respiratory symptoms include chest pain and hemoptysis.⁵ Patients may exhibit gastrointestinal (GI) symptoms, such as hematemesis or abdominal pain.¹ GPF can be complicated by sepsis, hydropneumothorax, or tension pneumothorax. Chest tube drainage containing food particles or bile has also been reported.⁴

Diagnosis of GPF requires a high index of suspicion and thorough investigations.⁴ The initial workup includes CXR, which may demonstrate pneumothorax, pleural effusion, or hydropneumothorax.⁴ An upper GI series, preferably with water-soluble contrast, may show contrast extravasation to the pleural cavity.⁶ CT with oral contrast may reveal contrast leakage into the pleural space or delineate the fistula tract.⁴ Moreover, upper GI endoscopy may identify the site of the fistula and can be utilized for therapeutic purposes.⁷ Occasionally, GPF is diagnosed intraoperatively.⁸

There are no standardized treatment protocols for GPF.⁷ Initial steps of management consist of treating GPF complications and providing an alternative to oral feeding (parenteral nutrition or jejunostomy feeding tube).^{5,7} Meanwhile, the closure of the fistula can be done endoscopically or surgically. Various endoscopic interventions have been used, including endoscopic clip stents, suturing, and fistula plug

application.^{7,9,10} Our patient was managed conservatively, and the fistula was closed endoscopically with an OTSC.

Over-the-scope clip is a relatively new endoscopic intervention that has been used for chronic fistulas with variable success rates ranging from 30% to 100%. Other indications for OTSC include GI hemorrhage and perforation.¹¹ The main determent for OTSC success is the indication for its application, which is higher in patients with perforation than in patients with fistula.¹ In addition, it is more effective in managing fistula secondary to SG compared with other distal GI fistulas. It is considered safe; however, stricture at the gastroesophageal junction and OTSC migration have been infrequently reported.² Our patient's surgical management (ie, attempt to close the fistula) failed, probably because of an ongoing leak that resulted in infection, abscess collection, and poor healing. Other surgical options include partial gastric resection, diaphragmatic repair, and washout and drainage of the chest.⁴ Roux-en-Y esophagojejunostomy has been used for chronic GPF.

In conclusion, pediatric GPF has been reported to have various causes (Table 1), but our patient is the first report of pediatric GPF secondary to SG. SG is not without its complications, and it must be reserved for morbidly obese patients in whom conservative management fails. In our case, endoscopic intervention with an OTSC resulted in fistula closure and patient improvement.

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CONFLICT OF INTEREST

None declared.

AUTHORS' CONTRIBUTIONS

DA: Literature review, discussion writing. MA: Writing the case description, discussion writing. NA: Literature review, discussion writing. HA: Editing and reviewing the case description and discussion. FA: Writing the case description. WA: Editing and reviewing the case description and discussion.

ETHICAL APPROVAL

The study was approved by the Ethical Committee (King Fahad Medical City, Riyadh, Saudi Arabia) and was conducted in accordance with the Helsinki Declaration.

CONSENT FOR PUBLICATION

The patient's parent was informed that data from the case would be submitted for publication and gave the written informed consent.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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