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Original Article

Rare and unique endoscopic retrograde cholangiopancreatography (ERCP) findings: insights from a 24-year single-center retrospective study

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ABSTRACT

Aim: Rare conditions causing biliary obstruction, such as Fasciola hepatica infestation, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and surgical clip migration, are infrequently encountered during ERCP. This study aims to analyze the clinical and demographic characteristics, diagnostic challenges, and management of these uncommon conditions in a single-center cohort.

Method: This retrospective study included 6,700 patients aged 18 and older who underwent ERCP between 2000 and 2024. Patients with common bile duct stones, benign fibrosis, or malignancy were excluded, resulting in 28 cases with rare biliary pathologies. Data on clinical presentations, imaging findings, and treatment outcomes were analyzed.

Results: Among the 6,700 patients, the incidence of Fasciola hepatica infestation was 0.06% (n = 4), SUMP syndrome 0.75% (n = 5), Lemmel syndrome 0.06% (n = 4), Wirsung duct stones 0.044% (n = 3), Mirizzi syndrome 0.15% (n = 10), and surgical clip migration 0.03% (n = 2). Cholangitis was the most common clinical presentation across most conditions, with notable differences in total bilirubin levels, common bile duct diameters, and associated imaging findings. All cases were successfully managed with ERCP, and no complications were observed.

Conclusion: This study underscores the importance of recognizing rare biliary pathologies in patients presenting with atypical symptoms or a history of biliary surgery. Although these conditions represent a small percentage of patients undergoing ERCP, their timely diagnosis and management are crucial to preventing complications. For this reason, we are presenting these rare conditions collectively in the literature for the benefit of endoscopists.

Keywords: endoscopic retrograde cholangiopancreatography, fasciola hepatica, sump syndrome, lemmel syndrome, clips migration

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Introduction

Endoscopic Retrograde Cholangiopancreatography (ERCP) is a complex but essential procedure with a wide range of indications. ERCP was first found in the 1960s as a diagnostic tool but its therapeutic abilities were rapidly realized (1). The most common indications for ERCP are choledocholithiasis, cholangitis, acute biliary pancreatitis, and unidentified obstructions in the biliary tree (2).

Choledocholololithiasis, which presents with jaundice and abdominal pain caused by obstruction of the common bile duct (CBD), is also encountered with interesting diagnoses other than benign fibrotic lesions. Fascioliasis is a zoonotic infection caused by Fasciola hepatica and is a rare pathology observed in ERCP, and there are studies reported in the literature. Apart from this, Mirzzi syndrome observed due to cholelithiasis and clip migration observed after laparoscopic cholecystectomy are also interesting ERCP findings observed in the literature. Another interesting diagnosis observed in ERCP after surgery is SUMP syndrome, which can be observed after bile duct surgery. Pancreaticolithiasis is one of the interesting diagnoses that we can diagnose and treat in ERCP without jaundice and obstruction in the bile ducts. Although these cases are presented as single cases or case series in the literature (2-5), we present them as a whole in this study based on a single center experience. In our study, we aimed to highlight the interesting and less commonly seen cases of ERCP findings. With this review of our experiences of ERCP, we hope to raise the clinical suspicion of physicians and share our experiences.

Methods

This observational retrospective study included 6,700 patients aged 18 years and older who underwent ERCP between 2000 and 2024 at the Endoscopy Center of Istanbul Training and Research Hospital. The medical reports and imaging findings of these cases were retrospectively reviewed using the hospital's medical record system to identify pathologies within the CBD.

Patients diagnosed with CBD stones/sludge, fibrosis, or malignancy were excluded from the study. The remaining cases, comprising rare or less commonly reported diagnoses and their prevalence, were analyzed in the context of international literature. The study received approval from the Ethics Committee of Istanbul Training and Research Hospital (Approval No: 2011-KAEK-50, Decision Date: January 10, 2025).

Based on the inclusion and exclusion criteria, 28 patients with unusual ERCP findings were selected for further analysis. These findings included rare conditions such as fascioliasis, SUMP syndrome, Lemmel syndrome, Wirsung calculus, Mirizzi syndrome, and bile duct clip migration. For these selected cases, demographic and clinical characteristics were recorded, including the most common presenting complaints at hospital admission, mean age, gender distribution, mean diameter of the CBD prior to ERCP, presence of a gallbladder, presence of gallbladder calculi, and mean total bilirubin levels prior to ERCP. The data were compiled and subjected to descriptive analysis.

Results

Of the 6,700 ERCP cases reviewed, 88.7% (n = 5,943) were diagnosed with choledocholithiasis, 4.2% (n = 281) with malignancy, and 6.01% (n = 94) with benign fibrosis; these cases were excluded from the study (Table 1). A total of 28 patients with rare or unusual pathologies were included in the study and categorized into six groups: Fasciola hepatica obstruction (n = 4; 0.06%), SUMP syndrome (n = 5; 0.75%), Lemmel syndrome (n = 4; 0.06%), Wirsung duct stones (n = 3; 0.044%), Mirizzi syndrome (n = 10; 0.15%), and clip migration (n = 2; 0.03%) (Table 1).

Table 1. Findings and Distribution of ERCP Diagnoses

Finding	Number	Percentage (%)
Calculi-Sludge	5943	88.7
Malignancy	281	4.2
Benign Fibrosis/Obstructive Lesion	94	6.01
Fasciola Hepatica	4	0.06
SUMP Syndrome	5	0.75
Clip Migration	2	0.03
Lemmel Syndrome	4	0.06
Mirizzi Syndrome	10	0.15
Wirsung Stone	3	0.04
TOTAL	6700	100

Table 2. Demographic and Clinical Characteristics of Patients with Rare ERCP Diagnoses

Cases	Total size	Age (mean)	Male (%)	Female (%)	HS (most)	CBD (mean, mm)	GS	TB (mg/dl)
Fasciola Hepatica	4	42.5	25	75	Cholangitis	14	-	9.6
SUMP Syndrome	5	57.8	20	80	Cholangitis	22	operated	3.6
Lemmel Syndrome	4	70.75	25	75	Abdominal pain	12	+	5.9
Wirsung Stone	3	63.3	100	0	Abdominal pain	6.6	+	1.06
Mirizzi Syndrome	10	47.4	70	30	Jaundice	6.5	+	2.7
Clip Migration	2	42.5	100	0	Abdominal pain	10	operated	4.7

HS: The complaint in hospital admission, CBD: Common bile duct diameter, GS: Gallbladder stone, TB: Total Bilirubin

Fasciola Hepatica Obstruction

Four patients (0.06%) were diagnosed with Fasciola hepatica infestation. The group included three females and one male, with a mean age of 42.5 years. The most common clinical presentation was cholangitis. The mean total bilirubin level was 9.6 mg/dL, and the mean diameter of the CBD was 14 mm. Ultrasound imaging revealed mobile and linear echogenic findings in two cases, and parasites were visualized during ERCP in all four cases (Table 2).

SUMP Syndrome

Five patients (0.75%) were diagnosed with SUMP syndrome. The mean age of this group was 57.8 years, consisting of four females and one male. All patients had a history of cholecystectomy and choledochoduodenostomy, and the most common presentation was cholangitis. The mean total bilirubin level was 3.6 mg/dL, and the mean CBD diameter was 22 mm (Table 2).

Lemmel Syndrome

Four patients (0.06%) were identified with Lemmel syndrome, with a mean age of 70.75 years. Three of these patients were female. The most common presentation was right upper quadrant (RUQ) pain. The mean total bilirubin level was 5.9 mg/dL, and the mean CBD diameter was 12 mm. All patients had cholelithiasis detected on pre-ERCP ultrasound, and three were diagnosed via MRCP before ERCP (Table 2, Figure 1).

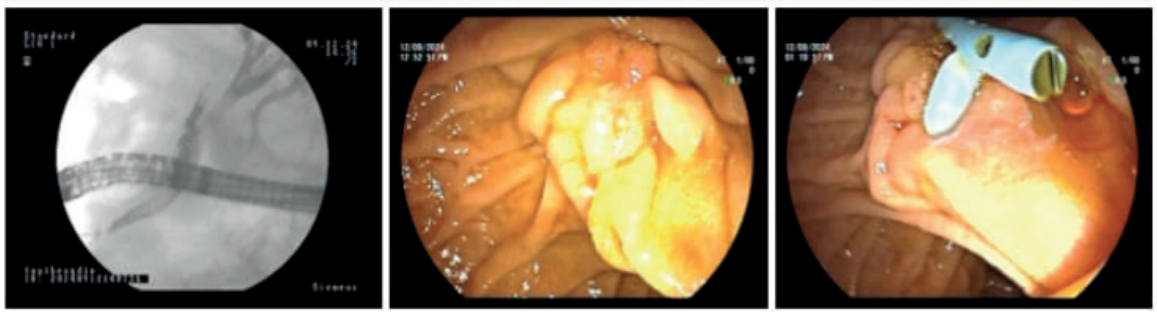

Figure 1. ERCP performed in a patient with Lemmel syndrome

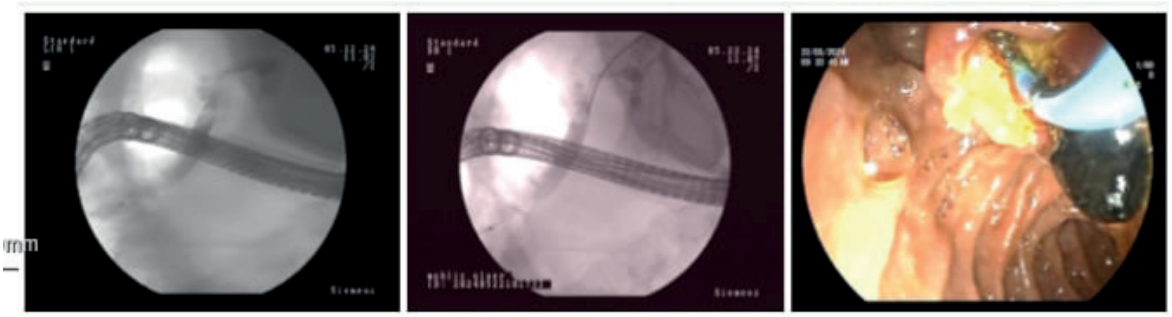

Figure 2. ERCP and stone extraction in a patient with Mirizzi syndrome

Wirsung Duct Stones

Three patients (0.044%) were found to have stones in the Wirsung duct, with a mean age of 63.3 years. All patients had a history of recurrent pancreatitis and presented with epigastric pain. The mean CBD diameter was 6.6 mm, and the mean total bilirubin level was 1.06 mg/dL. Pre-ERCP imaging (MRI and endoscopic ultrasound) revealed pancreatic duct dilation and pancreatic head stenosis, as well as cholelithiasis (Table 2).

Mirizzi Syndrome

Ten patients (0.15%) were diagnosed with Mirizzi syndrome, with a mean age of 47.4 years. Seven were male, and three were female. The most common clinical presentation was jaundice, with RUQ pain noted in half of the cases. Pre-ERCP imaging showed cholelithiasis in all cases, and one patient demonstrated pneumobilia. The mean total bilirubin level was 2.7 mg/dL, and the mean CBD diameter was 6.5 mm. Nine patients had type 1 Mirizzi syndrome, while one was classified as type 2 (Table 2, Figure 2).

Clip Migration

Two cases (0.03%) of clip migration were identified. Both patients were male, with a mean age of 42.5 years. The most common

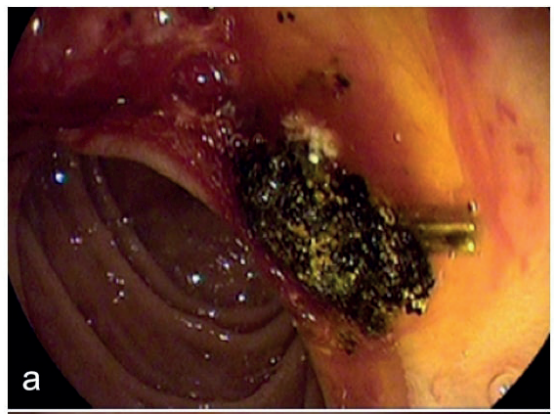

Figure 3. Stone extraction with ERCP in a patient with Mirizzi syndrome

presentation was RUQ pain. The mean CBD diameter was 10 mm, and the mean total bilirubin level was 4.7 mg/dL. One patient had undergone cholecystectomy 6 months prior, and the other 14 months prior. ERCP revealed bile duct sludge alongside migrated clips in both cases (Table 2, Figure 3).

Discussion

The findings of this study highlight the diversity and clinical significance of rare pathologies encountered during ERCP procedures. While the majority of cases involved common conditions such as choledocholithiasis (88.7%), malignancy (4.2%), and benign fibrosis (6.01%), this study focused on the less frequently observed diagnoses, comprising 0.42% (28 patients) of the total cohort. These included Fasciola hepatica obstruction, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and clip migration. The rarity of these conditions underscores the importance of maintaining a high index of suspicion, especially in patients presenting with atypical symptoms or inconclusive imaging findings. By systematically analyzing these rare cases, this study contributes valuable insights into their clinical presentations, diagnostic challenges, and management strategies, providing a foundation for improved recognition and treatment in clinical practice.

Fasciola hepatica is a rare trematode infection in which the parasites can cause obstruction in the biliary tract. Diagnosing fascioliasis during the biliary phase can be challenging due to its wide variety of clinical presentations. In our study, 4 out of 6,700 patients (0.06%) were found to have obstruction of the CBD caused by F. hepatica. ERCP was performed, and cholangiography revealed filling defects in the CBD. Following sphincterotomy, adult trematode flukes were identified as the cause of obstruction. Encountering F. hepatica flukes during ERCP is rare. Our findings align with the case review by Sharma et al., (1) who also reported successful sphincterotomy and fluke extraction, followed by CBD stenting. In their case, anaphylaxis occurred—a known but serious complication of parasitic infections. Similarly, Aghajanzadeh et al. described a case where F. hepatica eggs obstructed the CBD, and the patient underwent sphincterotomy and stenting, ultimately recovering without significant complications (3,4).

In contrast, Rinaldi et al. reported a case of F. hepatica presenting as a peripancreatic mass. Their patient exhibited elevated bilirubin levels, with ERCP revealing CBD stenosis and fibrosis (5). In our study, cholangitis emerged as the most common clinical presentation, consistent with other reports. However, our patients exhibited a higher mean total bilirubin level (9.6 mg/dL) compared to similar cases in the literature. Additionally, one of our patients presented with a unique ultrasonographic finding of linear, mobile lesions within the gallbladder, distinct from gallstones. The mean CBD diameter in our cases was 14 mm, which exceeds the average range of 8–12 mm typically observed in choledocholithiasis cases (6). Given that fascioliasis is endemic to specific regions, we recommend that all patients presenting with biliary obstruction or cholangitis should be queried about their travel history to endemic areas (2).

Lemmel syndrome is a rare condition characterized by obstruction and jaundice caused by a periampullary duodenal diverticulum. ERCP is often the primary treatment modality for this condition. In our study, 4 out of 6,700 patients (0.06%) were diagnosed with Lemmel syndrome. A periampullary diverticulum can lead to obstruction through several mechanisms, including direct compression of the CBD or ampulla of Vater, dysfunction of the sphincter of Oddi, or chronic fibrosis of the papilla (7). Despite its rarity, Lemmel syndrome is a critical differential diagnosis for patients presenting with obstructive jaundice.

The diagnosis can be made using imaging techniques such as CT or MRCP, but the gold standard remains lateral-view endoscopy

during ERCP (8). Goroztieta-Rosales et al. reported a similar case of Lemmel syndrome in a patient presenting with RUQ pain and elevated bilirubin levels. The diagnosis was confirmed via ERCP (7). Similarly, Bernshteyn et al. described an ERCP case where impacted food was identified within the diverticulum and successfully removed using a snare (9). The incidence of duodenal diverticula is reported to be approximately 11% in patients older than 70 years, but the exact incidence of Lemmel syndrome remains unknown. In our cohort of 6,700 ERCP cases, the incidence of Lemmel syndrome was 0.06%. The mean age of patients in our study was 70.75 years, consistent with findings in the literature. The majority of patients presented with RUQ pain and a mean CBD diameter of 12 mm, both of which align with previously reported clinical characteristics of Lemmel syndrome (6). These findings underscore the importance of considering Lemmel syndrome in elderly patients presenting with biliary obstruction, especially when a duodenal diverticulum is identified on imaging.

Pancreatolithiasis can occur in the Wirsung or Santorini ducts, the side branches of pancreatic ducts, or within the pancreatic parenchyma. MRCP and EUS are highly sensitive diagnostic tools for detecting pancreatolithiasis (10-13). In our study, all three cases of pancreatolithiasis involved male patients with a history of recurrent pancreatitis. The mean age of these patients was 63.3 years, and two had significant histories of alcohol use. The incidence of pancreatolithiasis in our cohort was 0.044%, reflecting the rarity of this condition. Notably, the mean CBD diameter and total bilirubin levels were within normal ranges in these cases, which is consistent with the absence of significant biliary obstruction.

Mirizzi syndrome is a rare cause of obstructive jaundice, resulting from a stone impacted in the cystic duct or neck of the gallbladder (14). According to the literature, the incidence of Mirizzi syndrome requiring surgical intervention ranges from 0.07% to 1.4% (15). In our cohort of 6,700 ERCP cases, the incidence of Mirizzi syndrome was notably lower, at 0.15%. Among our cases, one was classified as type 2 Mirizzi syndrome, while the remaining cases were type 1. These findings underscore the rarity of Mirizzi syndrome in clinical practice and highlight the need for a high index of suspicion in patients presenting with obstructive jaundice and relevant imaging findings.

Surgical clip migration into the CBD is one of the rarest causes of jaundice, with fewer than 100 cases reported in the literature (16). The most widely accepted theory suggests that chronic inflammation near the CBD leads to erosion, particularly in cases where the cystic duct is anatomically close to the CBD. Migration has been reported to occur up to 14 years post-cholecystectomy, although the majority of cases are documented within 2 years of surgery (17,18). Photi et al. reported a case of clip migration 16 years post-surgery, successfully managed with ERCP, similar to the cases in our study (19). In our series, all patients presented with RUQ pain and had a mean total bilirubin level of 4.7 mg/dL, approximately three times the upper normal limit. All cases were successfully treated with ERCP without any complications, highlighting the effectiveness of this approach in managing this rare condition.

SUMP syndrome is an exceptionally rare condition, primarily occurring in patients with a history of side-to-side choledochoduodenostomy (CDD) or open cholecystectomy.In SUMP syndrome, the portion of the CBD distal to the CDD becomes a reservoir for debris due to poor drainage, forming what is referred to as a “sump” (20,21). Abbarh et al., in their case series, reported three patients with SUMP syndrome, all with a history of CDD performed during open cholecystectomy. Two of their patients were successfully treated with a combination of percutaneous transhepatic cholangiography (PTC) and ERCP, while one patient did not return for the planned ERCP (21). In our study, both patients diagnosed with SUMP syndrome were successfully managed with ERCP alone, without the need for PTC. Similarly, Abraham

et al. described a patient with a history of CDD who presented with cholangitis and was treated with urgent ERCP (20).

Granata et al. reported a unique approach where they utilized an endoscopic overstitch device to reduce the diameter of the CDD, thereby preventing further accumulation of debris in the sump (22). In our study, cholangitis was the most common clinical presentation among patients with SUMP syndrome. The mean CBD diameter in this group was 22 mm, the largest observed in our study, reflecting the significant drainage dysfunction associated with this condition. All patients had a history of CDD, consistent with findings by Ağca et al., who reported SUMP syndrome in four cases of CDD and one case of choledochojejunostomy (23). These observations reinforce the association between prior biliary surgeries and the development of SUMP syndrome, emphasizing the importance of tailored treatment strategies in such cases.

The primary limitation of this study is its retrospective design, which inherently depends on the accuracy and completeness of recorded data. Additionally, the rarity of the conditions studied resulted in a small sample size for each subgroup, limiting the generalizability of the findings. The study was conducted at a single center, which may not reflect variations in patient demographics or clinical practices across different regions. Finally, the lack of long-term follow-up data for the treated patients precludes an analysis of recurrence rates or long-term outcomes.

Conclusion

This study highlights the importance of recognizing rare and unusual causes of biliary obstruction, such as Fasciola hepatica infestation, SUMP syndrome, Lemmel syndrome, Wirsung duct stones, Mirizzi syndrome, and surgical clip migration. Although these conditions represent a small percentage of patients undergoing ERCP, their timely diagnosis and management are crucial to preventing complications. Advanced imaging techniques, such as MRCP and EUS, alongside ERCP, play a vital role in the accurate diagnosis and treatment of these rare pathologies. Our findings emphasize the need for a high index of suspicion, particularly in patients with atypical presentations or relevant surgical histories. Future studies with larger cohorts and prospective designs are needed to further explore these rare conditions, improve diagnostic accuracy, and optimize treatment strategies.

Ethical approval

The study was performed according to the Helsinki Declaration. The study protocol was approval by the Ethics Committee of Istanbul Training and Research Hospital (Approval No: 2011-KAEK-50, Decision Date: January 10, 2025).

Author contribution

The authors confirm contribution to the paper as follows: Study conception and design: EÇ, OOK, MMS; data collection: EÇ, OOK, SZK, MAD, BD; analysis and interpretation of results: EÇ, OOK, SZK, MAD, BD; draft manuscript preparation: EÇ, OOK, MMS. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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Original Article

Pilonidal disease management results

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ABSTRACT

Objective: This study aimed to compare the effectiveness, recurrence, and complication rates of different surgical methods used in the treatment of pilonidal sinus disease.

Methods: A total of 49 patients who underwent elective surgery between September 2023 and September 2024 were retrospectively evaluated. Surgical techniques included unroofing, liquid phenol application, Limberg flap, and primary closure. The primary endpoint was recurrence; the secondary endpoint was complication rate.

Results: Of the 49 patients, 41 were male and 8 were female. The most commonly performed procedure was unroofing (51.1%). The overall complication rate was 8.4% and recurrence rate was 4.2%. There was no statistically significant difference between surgical methods in terms of recurrence or complications ($p > 0.05$).

Conclusion: Each surgical method may be effective for selected patient groups. Treatment should be individualized based on patient characteristics and surgeon experience. Larger-scale studies with longer follow-up are needed.

Keywords: Pilonidal disease, recurrence, complications, Limberg flap, unroofing, phenol treatment.

Introduction

Pilonidal disease is an infection of the subcutaneous tissue in the gluteal cleft. Acute pilonidal disease may present as an abscess characterised by pain, swelling and redness in the coccyx or as a pilonidal sinus characterised by pain, swelling and sinuses in the chronic period. It is usually seen in men in their twenties. It is more common in hairy men. Obesity, people who drive sitting for a long time, sedentary life, development of furuncles in other parts of the body are also risk factors for pilonidal disease (1).

Discussions about the pathogenesis of pilonidal disease development continue. Although it is historically thought to be embryological development, it is thought that the development of pilonidal disease is a secondary process with the studies of Karydakis and Bascom (2,3). Theories converge on the idea that foreign bodies such as hair, hair, feathers accumulate in the natal cleft and cause inflammation (4).

There is no gold standard method in the treatment of pilonidal disease and various surgical procedures can be applied. Leaving the sinus to secondary healing after primary

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excision, unroofing, Bascom Cleft Procedure, Karydakis Flap, Limberg flap application, phenol application, laser applications are among the preferred methods in current applications (4). In our study, we aimed to evaluate the results of elective pilonidal sinus applications performed in our clinic in the last year.

Material and Method

This study was approved by the institutional ethics committee and informed consent was obtained from all participants. Patients who underwent elective operation by a single surgeon for pilonidal sinus between September 2023 and September 2024 in our clinic were retrospectively evaluated. Patients older than 18 years were included in the study. Patients who underwent abscess drainage due to pilonidal sinus abscess were excluded from the study. Patients who underwent unroofing, primary closure after excision, secondary healing after excision, Limberg flap procedure, liquid phenol due to pilonidal disease were included in the study. The study's primary endpoint is the recurrence rates during the follow-up period, and the secondary endpoint is the complication rates of the procedures.

Patient data were analysed by examining outpatient clinic records and operation notes. The data obtained were transferred to the database prepared in Microsoft Excel programme and analysed with Jamovi programme. After descriptive statistics, normality was evaluated by Kolmogrov-Smirnow test in the comparison of groups, Independent T-Test was applied in the comparison of paired groups with normal distribution, and Mann-Whitney-U test was applied in the comparison of paired groups without normal distribution. In the comparison of two or more groups, One Way Anova test was applied for data showing normal distribution and Kruskall-Wallis test was applied for tests that did not show normal distribution. In our study, the confidence interval was accepted as 95% and 0.05 was accepted as the limit for the significance of p value.

Results

A total of 49 patients, 8 females and 41 males, were included in the study. The mean age of the patients was 24.3 ± 5.5 years. Seven (14.9%) of the patients included in the study were recurrence cases. In 25 patients (51.1%) unroofing, in 14 patients (27.7%) liquid phenol, in 9 patients (19.1%) Limberg flap, in 1 patient (2.1%) primary closure after excision was performed (Table 1). 4 patients (8.5%) developed complications secondary to the procedure. One patient with unroofing developed postspinal headache, two patients with liquid phenol developed skin burns outside the procedure area. One patient who was performed Limberg flap operation developed ischaemia at the lower end of the flap. In the patient who underwent Limberg flap, recurrent pilonidal disease which developed in the area of ischaemia at the 6th month of follow-up, unroofing was applied to this patient, the skin healed and no recurrence was detected at the 3rd month control after treatment. One patient who underwent unroofing developed recurrence at the 11th month of follow-up, liquid phenol was applied to this patient and no recurrence occurred after treatment. The mean follow-up period of the patients was 10.5 ± 2.8 months. There was no statistically significant difference between the surgical procedure and recurrence (p:0,843). There was no statistically significant difference between the surgical procedure and the development of complications (p: 0,051) (Table 2).

Table 1. Surgical Procedures Performed

Surgical Procedure	Number	%
Unroofing	25	51,1
Liquid Phenol Application	14	27,7
Limberg Flap Procedure	9	19,1
Primary Closure	1	2,1
Total	49	100

Table 2. Surgical Procedures and Outcomes

Surgical Procedure	Recurrence n, (%)	P value	Complication n, (%)	P value
Unroofing	1 (%2,1)	0,843	1 (%2,1)	0,051
Liquid Phenol Application	-		2 (%4,2)	
Limberg Flap Procedure	1 (%2,1)		1 (%2,1)	
Primary Closure	-		-	
Total	2			

Discussion

The effectiveness and complication rates of surgical methods applied in the treatment of pilonidal disease are frequently discussed in the literature. In our study, the results of different surgical methods applied in our clinic were evaluated retrospectively. The results obtained are largely consistent with the existing literature.

The unroofing method, which was preferred in the majority of our patients (51.1%), is widely recommended and accepted as a minimally invasive method in the literature. The low recurrence rates obtained with the unroofing method support the effectiveness of the method. However, postspinal headache was observed in one patient, indicating that this method may lead to complications, albeit rare. It is also reported in the literature that unroofing is a reliable method with low complication rates (5-8).

The observation of complications such as skin burns in patients treated with liquid phenol reveals the potential risks of this method. Although phenol is an effective treatment option, it is clear that contact with the skin should be carefully controlled during the procedure (9,10). The cases of flap ischaemia and recurrence in patients treated with the Limberg flap method show that there is a risk of complications and recurrence in more invasive methods. The fact that the patient with recurrence was treated with unroofing and a successful result shows that minimally invasive methods can be effective even in cases of recurrence (11).

In our study, there was no statistically significant difference ($p > 0.05$) between the surgical procedures performed and the development of complications and recurrence rates, indicating that each method may offer advantages and disadvantages according to its own patient group. This emphasises that the choice of treatment should be individualised according to patient characteristics, stage of the disease and the experience of the surgeon.

This study has certain limitations. The retrospective design, relatively small sample size—especially in some surgical subgroups—and the short follow-up period may reduce the generalisability of the findings. Additionally, lack of standardized criteria for recurrence and complication definitions may affect outcome interpretation. Prospective, multicentre studies with longer follow-up are needed to validate these findings

Conclusion

In conclusion, surgical methods applied in our clinic were found to be generally effective and safe in the treatment of pilonidal disease. However, the adoption of individualised patient-specific approaches in the selection of methods is critical in reducing complications and recurrence rates. Future large-scale, prospective and long follow-up studies will increase the knowledge in this field.

Ethical approval

This study was approved by the institutional

ethics committee (Decision number: 2025/447, date: 30.04.2025) and informed consent was obtained from all participants.

Author contribution

The author confirm contribution to the paper as follows: Study conception and design: CU; data collection: CU; analysis and interpretation of results: CU; draft manuscript preparation: CU. The author reviewed the results and approved the final version of the manuscript.

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Original Article

Diagnostic performance of ChatGPT in detecting gastrointestinal tract perforation on chest radiographs: a comparative study

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ABSTRACT

Background: Gastrointestinal (GI) tract perforation is a surgical emergency requiring rapid diagnosis, often via chest radiography. Artificial intelligence (AI), including large language models like ChatGPT, has potential to enhance medical imaging but its efficacy in detecting GI perforation is unclear. We compared the diagnostic accuracy of ChatGPT 3.5 and 4 with human experts in interpreting chest radiographs for GI perforation.

Methods: This retrospective study, approved by the Arel University Hospital Ethics Committee (E-52857131-050.06.04-455896), analyzed 504 chest radiographs from patients diagnosed with GI perforation between 2010 and 2021. Radiographs were classified into three groups: definite GI perforation, suspicious requiring further imaging, or no perforation. Two clinicians (emergency medicine specialist and general surgeon) independently evaluated radiographs, followed by ChatGPT 3.5 and 4 using a standardized prompt. Diagnostic accuracy was assessed with chi-square tests, and decision-making times with Student's t-test ($p<0.05$ for significance).

Results: Of 504 patients (11.1% female, mean age 45.4 years), human evaluators correctly classified 80.1% of radiographs, compared with 3.9% for ChatGPT 3.5 and 5.9% for ChatGPT 4 ($p<0.001$). ChatGPT models were faster ($p<0.001$) but failed to interpret 94.1–96.1% of radiographs, often recommending clinical consultation.

Conclusion: General-purpose ChatGPT models lack the accuracy for reliable GI perforation diagnosis on chest radiographs. Specialized AI models, trained on medical imaging datasets, are needed to improve diagnostic precision and support clinical workflows.

Keywords: AI in healthcare, artificial intelligence, emergency radiology, radiology AI

Introduction

Gastrointestinal (GI) tract perforation is a life-threatening condition necessitating urgent surgical intervention. Chest radiography, widely used to detect pneumoperitoneum, is limited by high workloads and human error, which can delay diagnosis. GI perforations, such as those caused by peptic ulcers, appendicitis, or diverticulitis, carry significant morbidity and mortality if not diagnosed promptly, with mortality rates ranging from 10–30% depending on the underlying cause and time to intervention (1). In busy emergency departments, where clinicians often face high patient volumes and time constraints, rapid and accurate diagnosis is critical to

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improving patient outcomes. The complexity of interpreting subtle radiological signs, such as free intraperitoneal air, underscores the need for tools that can enhance diagnostic efficiency without compromising accuracy.

Artificial intelligence (AI), particularly large language models like ChatGPT (OpenAI), has shown promise in medical imaging by reducing diagnostic time and errors. ChatGPT, developed by OpenAI, is a conversational AI model based on the GPT architecture, designed to process and generate human-like text and, in later versions, interpret visual inputs such as images. Since its release, ChatGPT has been adopted globally across diverse fields, including education, customer service, content creation, and healthcare, due to its ability to process vast datasets and provide contextually relevant responses (2,3). In healthcare, its applications range from answering medical queries to assisting with clinical documentation, but its role in diagnostic imaging remains underexplored. This study evaluates the diagnostic performance of ChatGPT 3.5 and 4 in detecting GI perforation on chest radiographs compared with human experts, assessing accuracy, speed, and clinical applicability.

Methods

Study Design and Participants

This retrospective study, approved by the Arel University Hospital Non-Invasive Ethics Committee (E-52857131-050.06.04-455896), included 504 patients diagnosed with GI perforation between January 2010 and December 2021 at Arel University Hospital. Patients were identified from emergency department records, and posteroanterior chest radiographs were retrieved. Informed consent was obtained for image use in research.

Radiographs were classified into three groups:

1. Definite GI perforation (no further imaging needed).

2. Suspicious for GI perforation (additional imaging required).

3. No GI perforation (further imaging mandatory) (Figure 1).

Human Evaluation

Two experienced clinicians (an emergency medicine specialist and a general surgeon) independently classified radiographs without access to ChatGPT outputs or each other's assessments. A consensus diagnosis was established for discordant cases via discussion.

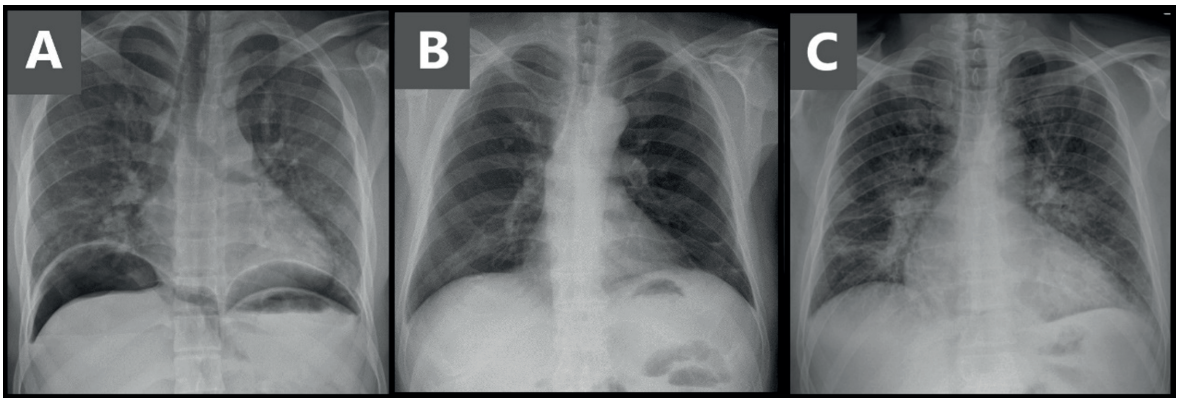

Figure 1. Cheast graphs were divided into 3 groups: a) GI perforation is certain; b) GI perforation is suspicious; c) no GI perforation

AI Evaluation

Radiographs were uploaded to ChatGPT 3.5 and 4 using a single authenticated account. The standardized prompt was: "Examine the chest radiograph and identify any pneumoperitoneum." Each image was input once without modification to ensure consistency. ChatGPT outputs were categorized into the three groups based on their responses or lack thereof (e.g., inability to interpret) (Table 1).

Statistical Analysis

Diagnostic accuracy was calculated as the proportion of correct classifications compared with the human consensus. Chi-square tests assessed differences in correct classification rates ($p<0.05$ for significance). Decision-making times were compared using Student's t-test ($p<0.001$). Confidence intervals (95% CI) were calculated for accuracy estimates, and Cohen's d was used for time comparisons. Analyses were conducted using SPSS version 25.0 (IBM Corp., Armonk, NY, USA).

Results

Patient Characteristics

Of 504 patients, 56 (11.1%) were female, and the mean age was 45.4 years (range 18–86). All patients had confirmed GI perforation via chest radiography (n=394, 78.2%), abdominal tomography (n=94, 18.7%), or diagnostic laparotomy (n=16, 3.2%).

Human Evaluation

Initial human evaluation classified 394 (78.2%) radiographs as definite GI perforation (Group 1), 94 (18.7%) as suspicious (Group 2), and 16 (3.2%) as no perforation (Group 3). Secondary review, conducted without time constraints, reclassified 413 (81.9%; 95% CI 78.3–85.2) to Group 1, 77 (15.3%; 95% CI 12.2–18.8) to Group 2, and 14 (2.8%; 95% CI 1.5–4.6) to Group 3.

AI Evaluation

ChatGPT 3.5 correctly classified 3.9% (20/504; 95% CI 2.4–6.0) of radiographs, and ChatGPT 4 classified 5.9% (30/504; 95% CI 4.0–8.4) correctly ($p<0.001$ vs. human evaluation). Both models failed to interpret 94.1–96.1% of radiographs, often responding with "consult a healthcare professional." Decision-making times were significantly faster for ChatGPT (mean 2.3 s for 3.5, 2.1 s for 4) than humans (mean 45.6 s; $p<0.001$, Cohen's $d=3.2$).

Statistical Findings

Chi-square tests confirmed that human classifications were significantly more accurate than ChatGPT ($\chi^2=392.4$, $p<0.001$). Time efficiency analysis showed ChatGPT's speed advantage ($t=28.7$, $p<0.001$) (Table 2).

Table 1. Diagnostic Accuracy and Time Efficiency of Human Experts and ChatGPT for GI Perforation on Chest Radiographs

Evaluator	Group 1: Definite GI Perforation (n, % [95% CI])*	Group 2: Suspected GI Perforation (n, % [95% CI])*	Group 3: No Perforation (n, % [95% CI])*	Non-Interpretable Rate (n, %)	Total Time (s)	Time Range (s)
Human (ER)	394 (78.2, 74.3–81.7)	94 (18.7, 15.3–22.4)	16 (3.2, 1.8–5.1)	0 (0.0)	10,206	60–720
Human (Home)	413 (81.9, 78.3–85.2)	77 (15.3, 12.2–18.8)	14 (2.8, 1.5–4.6)	0 (0.0)	14,952	60–780
ChatGPT 3.5	20 (4.0, 2.4–6.0)	0 (0.0, 0.0–0.7)	0 (0.0, 0.0–0.7)	484 (96.0)	1,159	48–432
ChatGPT 4	30 (6.0, 4.0–8.4)	0 (0.0, 0.0–0.7)	0 (0.0, 0.0–0.7)	474 (94.0)	1,075	48–420

Table 2. Correct Diagnoses by Human Experts and ChatGPT for GI Perforation on Chest Radiographs

Evaluator	Correct Diagnoses (n/504)	Accuracy (% [95% CI])	Non-Interpretable Rate (n, %)
Human (ER)	394	78.2 (74.3–81.7)	0 (0.0)
Human (Home)	413	81.9 (78.3–85.2)	0 (0.0)
ChatGPT 3.5	20	4.0 (2.4–6.0)	484 (96.0)
ChatGPT 4	30	6.0 (4.0–8.4)	474 (94.0)

Discussion

This study highlights the limitations of general-purpose ChatGPT models (3.5 and 4) in diagnosing GI perforation from chest radiographs, with accuracy rates of 3.9% and 5.9%, respectively, compared with 80.1% for human evaluators. The poor performance likely stems from the models' lack of specific training for medical imaging, as they are designed for general text and image processing rather than radiological interpretation. Specialized AI models, trained on curated medical datasets, have demonstrated superior performance in other imaging tasks, such as detecting pneumothorax or lung cancer, suggesting potential for improvement with tailored algorithms (4,5).

ChatGPT's speed advantage (2.1–2.3 s vs. 45.6 s) is notable but clinically irrelevant given its low accuracy. The standardized prompt used may have limited performance, as more specific or iterative prompts could enhance output quality. Additionally, the models' frequent inability to interpret radiographs and default to recommending clinical consultation underscores their unsuitability for standalone diagnostic use.

In medicine, ChatGPT has been explored in various applications beyond imaging, including clinical decision support, patient education, and medical documentation. For instance, studies have investigated its ability to answer medical queries, assist in drafting clinical notes, and even support medical education by generating practice questions or summarizing complex literature (2,6,7). In radiology, ChatGPT has been tested for tasks such as generating radiology reports and interpreting imaging findings, though with mixed results (8,9). A systematic review by Keshavarz et al. found that while ChatGPT shows promise in radiology report generation, its diagnostic accuracy for complex imaging tasks, such as identifying subtle abnormalities, remains limited due to insufficient training on specialized datasets (7). Similarly, Ahyad et al. reported that ChatGPT could reduce reporting time for radiologists but struggled with nuanced interpretations, aligning with our findings (8).

Literature involving ChatGPT in medical contexts highlights both its potential and limitations. For example, Xue et al. noted its utility in translational medicine for summarizing research findings but cautioned against overreliance due to potential inaccuracies (2). Fijačko et al. demonstrated that ChatGPT could pass certain medical exams, suggesting competence in knowledge-based tasks, but its performance in practical, image-based diagnostics remains inadequate (5). These studies collectively emphasize the need for specialized AI training to bridge the gap between general-purpose models and clinical applications. Our findings align with this, as ChatGPT's inability to reliably detect GI perforation underscores the necessity for domain-specific AI models in radiology (10).

Limitations include the use of general-purpose ChatGPT models, which are not optimized for radiology, and the retrospective design, which may not reflect real-time clinical challenges. Future research should explore fine-tuned AI models, incorporate diverse imaging modalities (e.g., CT), and assess iterative prompting strategies. Additionally, integrating ChatGPT with other AI tools, such as computer-aided

detection systems, could enhance its utility in clinical workflows (4). Ethical concerns, including data privacy and algorithmic bias, must also be addressed to ensure safe AI integration into clinical practice (11,12).

Conclusion

General-purpose ChatGPT models are not suitable for diagnosing GI perforation on chest radiographs due to low accuracy, despite faster processing times. Specialized AI models, developed with medical imaging expertise, are needed to enhance diagnostic precision and support clinicians. Addressing ethical and technical challenges will be crucial for AI's safe integration into medical workflows.

Ethical approval

The study was approved by Arel University Hospital Ethics Committee (number: E-52857131-050.06.04-455896).

Author contribution

The authors confirm contribution to the paper as follows: Study conception and design: MT, OT; data collection: MT, OT; analysis and interpretation of results: MT, OT; draft manuscript preparation: MT, OT. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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Original Article

Use of seton in chronic pilonidal sinus disease: a retrospective evaluation

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ABSTRACT

Objective: To evaluate the efficacy, complication rates, recurrence, infection, and need for reoperation of the seton technique in the treatment of chronic pilonidal sinus disease.

Methods: A retrospective review was conducted using our hospital's general surgery database, analyzing patients treated with the seton technique for chronic pilonidal sinus between December 2018 and January 2020. Patients with incomplete records were contacted by phone for verification and included in the study. A total of 30 patients (6 female [20%], 24 male [80%]) were evaluated. The mean age was 26.4 ± 5.2 years (range: 18–39). Seton duration ranged from 16 to 24 weeks (mean: 20 weeks). In three patients (10%), wound closure was not achieved; these cases required skin incision, seton removal, and phenol injection support.

Results: The technique was used in non-complicated cases (>2 cm size, <2 orifices) without a history of recurrence. This minimally invasive approach offered advantages such as low pain, no need for dressing or drainage, >90% cost reduction, and no hospital stay. Additional intervention was required in three cases (10%); the recurrence rate was 6.7%, and the infection rate was 3.3%.

Conclusion: The seton technique is an effective, minimally invasive method for chronic pilonidal sinus disease, similar to fistula treatment principles. Phenol support is recommended to reduce long-term recurrence rates.

Keywords: Pilonidal sinus, seton technique, minimally invasive surgery, recurrence.

Introduction

Chronic pilonidal sinus disease, commonly known as pilonidal disease, is a frequent inflammatory condition in the sacrococcygeal region, more prevalent in young males. The disease is characterized by sinus tract formation due to hair and debris accumulation, presenting with acute abscess, chronic discharge, or recurrent episodes. Standard treatments, such as excision with primary closure, open wound healing, or flap techniques, are associated with high recurrence rates (10–30%), prolonged healing times, and postoperative morbidity (1). In recent years, minimally invasive approaches, particularly the seton technique adapted from fistula-in-ano treatment, have shown promising results in pilonidal sinus management (2). The seton facilitates gradual drainage and fibrosis, promoting healing while offering advantages such as minimal incision, reduced pain, no need

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for dressing or drainage, cost-effectiveness, and improved patient comfort (3).

This study aims to retrospectively evaluate the efficacy, complications, and patient outcomes of the seton technique for chronic pilonidal sinus disease in our general surgery clinic. The technique was indicated for non-complicated cases (>2 cm size, <2 orifices) without prior recurrence.

Materials and Methods

Study design and patient selection

This retrospective cohort study was conducted using data from our hospital's general surgery database, covering the period from December 2018 to January 2020. Patients diagnosed with chronic pilonidal sinus and treated with the seton technique were screened. Inclusion criteria were non-complicated chronic pilonidal sinus (>2 cm sinus size, <2 orifices), no history of recurrence, and age between 18 and 40 years. Exclusion criteria included complicated cases (abscess, fistulization), recurrent cases, and inadequate follow-up data.

A total of 30 patients were included (6 female [20%], 24 male [80%]). The mean age was 26.4 ± 5.2 years (range: 18–39). For cases with incomplete records, patients were contacted by phone to verify data; unverified cases were excluded. Ethical approval was obtained from the Istanbul Medipol University Non-Interventional Ethics Committee (approval number: E-10840098-772.02.6312). Due to the retrospective nature of the study, patient consent was waived.

Surgical technique

The seton technique, adapted from fistula treatment principles, involved a minimal pit mouth incision (0.5–1 cm) and placement of a 1-0 polypropylene suture in the sinus tract. The seton duration ranged from 16 to 24 weeks (mean: 20 weeks). Weekly outpatient follow-ups included gradual tightening or replacement of the seton. No drainage or dressing was required. In three patients (10%), wound closure was not achieved by week 24; these cases underwent skin incision, seton removal, and 80% phenol injection to support granulation (4).

Evaluation parameters

Patients were assessed for recurrence (need for reoperation), infection (postoperative abscess/discharge), wound healing time, pain score (VAS 0–10), hospital stay duration, analgesia requirements, patient satisfaction, and cost. The mean follow-up duration was 12 months (range: 6–24 months). Data were analyzed using SPSS 25.0, with descriptive statistics (mean ± SD) reported.

Results

Patient demographics are summarized in Table 1. All cases were non-complicated, with a mean sinus size of 3.2 ± 0.8 cm and a mean of 1.4 ± 0.5 orifices.

Complications and outcomes

Wound Healing: Complete closure was achieved in 27 patients (90%) by week 24. Three patients (10%) required additional intervention (skin incision + phenol), with healing delayed by 8–12 weeks.

Recurrence: Two patients (6.7%) experienced recurrence at 6 months, treated with repeat seton placement.

Infection: One patient (3.3%) had mild discharge, resolved with antibiotics. No major abscesses were observed.

Table 1. Patient demographics

Parameter	Value (n=30)
Gender (Female/Male)	6/24 (20%/80%)
Age (mean ± SD)	26.4 ± 5.2 years
Seton Duration (mean)	20 ± 2.5 weeks
Follow-up Duration (mean)	12 ± 4 months

Table 2. Seton vs. standard excision comparison (literature-based)

Feature	Seton Technique (This Study)	Standard Excision (1)
Incision Size	Minimal (<1 cm)	Wide (3–5 cm)
Pain (VAS)	2.1 ± 1.2	4.5 ± 2.0
Dressing Requirement	None	Yes (weekly)
Drainage Tube	None	Yes (70%)
Cost	Very Low (>90% savings)	High
Comfort/Patient Satisfaction	High (8.7/10)	Moderate (6.5/10)
Postoperative Analgesia	Minimal (<1 week, 73%)	Prolonged (1–2 weeks, 60%)
Hospital Stay	None (0 days)	3–5 days
Recurrence Rate	6.7%	20–30%

Pain and Comfort: Mean VAS score was 2.1 ± 1.2, attributed to minimal incision. Postoperative analgesia was unnecessary or lasted <1 week in 73% of patients. No drainage tubes or dressings were used.

Hospital Stay and Cost: No hospital stay was required (outpatient surgery). Costs were >90% lower than standard excision techniques due to reduced material use and no hospitalization.

Patient Satisfaction: Mean satisfaction score was 8.7/10, with 93% of patients resuming daily activities within 1 week.

The advantages of the seton technique (similar to fistula treatment) include minimal incision, low pain, no dressing/drainage, >90% cost reduction, high comfort, minimal analgesia, and no hospital stay, as compared to standard excision in Table 2.

Discussion

The seton technique is a promising minimally invasive alternative for pilonidal sinus treatment. Adapted from fistula-in-ano management, it eliminates the sinus tract through gradual drainage and fibrosis, reducing recurrence (2). Our study demonstrated high success (90% closure), though three cases required phenol support, consistent with conservative approaches in the literature (4). The recurrence rate (6.7%) was below the literature average (10–30%), likely due to early intervention and diligent follow-up via phone (5).

Limitations include the retrospective design and small sample size (n=30). Phone verification mitigated data gaps but introduced potential bias. Future studies should employ prospective, randomized controlled designs. The technique's advantages, particularly in young patients (mean age: 26 years), support rapid socioeconomic recovery.

Conclusion

The seton technique is an effective, minimally invasive method for chronic pilonidal sinus disease with minimal morbidity and high patient satisfaction. It is recommended for non-complicated cases, with adjuvant treatments like phenol to reduce recurrence risk. This approach offers superior comfort and cost advantages over traditional surgery.

Ethical approval

The study was approved by Istanbul Medipol University Non-Interventional Ethics Committee (Number: E-10840098-772.02.6312).

Author contribution

The authors confirm contribution to the paper as follows: Study conception and design: MT, AG; data collection: UUŞ; analysis and interpretation of results: MT, UUŞ; draft manuscript preparation: MT, AG, UUŞ. All authors reviewed the results and approved the final version of the article.

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Conflict of interest

The authors declare that there is no conflict of interest.

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Case Report

A rare cause of small bowel obstruction internal herniation due to Allen-masters Syndrome: case report and literature review

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ABSTRACT

Background: Internal hernias are rare and constitute only a small fraction of bowel obstruction cases. Herniation through the broad ligament of the uterus, known as Allen-Masters syndrome, is an exceptionally rare cause of small bowel obstruction.

Case Report: We present the case of a 56-years-old woman with clinical signs indicative of mechanical bowel obstruction. During diagnostic laparoscopy, a segment of the small bowel was found entrapped within the broad ligament (ligamentum latum uteri). The entrapped bowel loop was released, and the ligament defect was sutured.

Conclusion: A defect in the ligamentum latum uteri, as seen in Allen-Masters syndrome, is a rare and often incidental finding in female patients presenting with ileus. This syndrome may account for nonspecific symptoms such as dyspareunia, dysmenorrhea, and both acute and chronic pelvic pain. Allen-Masters syndrome can be effectively diagnosed and treated through a laparoscopic approach.

Keywords: Allen-Masters syndrome; Broad ligament; internal hernia; intestinal obstruction; small bowel herniation.

Introduction

Bowel obstruction arises when the normal passage of intraluminal contents is interrupted. Approximately 80% of mechanical intestinal obstructions involve the small bowel (1,2). In developed countries, adhesions are the most frequent cause, followed by hernias, malignancies, and various infectious and inflammatory conditions. Acute mechanical small bowel obstruction is a common clinical scenario that necessitates prompt surgical evaluation (3). It is responsible for 2-4% of emergency department visits, approximately 15% of hospital admissions, and 20% of emergency abdominal surgeries (4,5).

Herniation through a defect in the broad ligament was first reported during an autopsy series (6,7). The clinical syndrome associated with a defect in the broad ligament, typically following trauma during delivery, was first

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described by Allen and Masters (8). This syndrome is characterized by symptoms such as dyspareunia, excessive physical fatigue, and dysmenorrhea. In 1955, Allen and Masters established a connection between traumatic delivery and lacerations in the posterior leaf of the broad ligament, thus defining this condition (8). Patients commonly present with persistent pelvic pain, dyspareunia, and menstrual irregularities. Herniation through the broad ligament of the uterus represents a rare form of internal hernia, accounting for approximately 4–7% of all cases (9).

This study aims to report a case of acute small bowel obstruction resulting from Allen-Masters Syndrome. Diagnostic and therapeutic considerations are discussed, accompanied by a comprehensive literature review.

Case Presentation

Patient Information: A 56-years-old female patient presented to the emergency department on September 20, 2024, with complaints of abdominal pain, nausea, and vomiting that had persisted for three days. Her medical history includes diagnoses of diabetes mellitus, hypertension, and hyperlipidemia, and she underwent cataract surgery approximately one month ago. She has a history of two cesarean sections as intra-abdominal surgeries. Her BMI was calculated as 23,14 kg/m^2 with no history of smoking, alcohol use, or known allergies. Laboratory parameters at admission showed hemoglobin (Hgb) 13.8 g/dL, white blood cell count (WBC) 10.59 x10^3/µL, pH 7.40, lactate 2.2 mmol/L, C-reactive protein (CRP) 18.3 mg/L, creatinine 2.65 mg/dL, urea 157.3 mg/dL, sodium 129 mmol/L, and glucose 109 mg/dL.

Clinical Findings: On clinical examination, dryness of the mucous membranes and darkened urine were observed. Abdominal examination revealed distension and tenderness, particularly in the lower quadrants. Drainage consistent with intestinal content was noted from the nasogastric tube.

Diagnostic Assessment: Abdominal computed tomography (CT) revealed air-fluid levels in the small intestine with dilation, reaching up to 37 mm at its widest point (Figure 1). The transition zone of the dilation was observed in the pelvic region, leading to a preliminary diagnosis of small bowel obstruction, and the patient was taken to surgery.

Therapeutic Interventions: The patient underwent abdominal exploration via diagnostic laparoscopy. During the exploration, a 5 cm segment of ileal loop approximately 40 cm proximal to the ileocecal valve was found to have herniated and become trapped within a defect in the broad ligament adjacent to the uterus on the left side (Figure 2). The trapped ileal segment was released laparoscopically. Examination of the defect revealed a peritoneal opening located between the round ligament and fallopian tube, consistent with Type 1 Allen-Masters syndrome (Figure 3). No signs of ischemia were observed in the small bowel, and no additional surgical intervention was deemed necessary. The peritoneal defect was repaired intracorporeally using 3-0 polyglactin 910 sutures, and the procedure was completed without complications.

Follow-up and Outcome of Interventions: Postoperatively, the patient was monitored with nasogastric drainage tube for one day. Oral intake was initiated on postoperative day

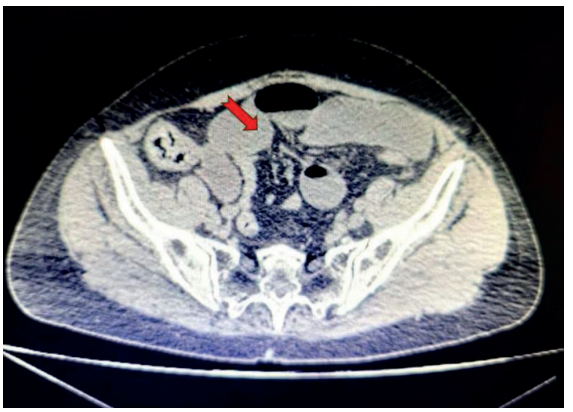

Figure 1. Abdominal CT scan showing dilated small intestine loops in the pelvic region and a transition zone indicating luminal narrowing in the ileal loops

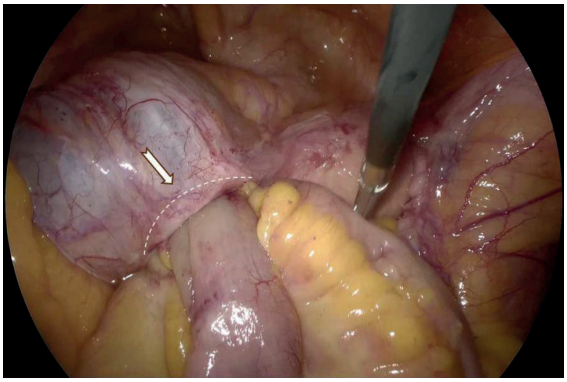

Figure 2. Compressed ileal loop herniating through a defect in the broad ligament

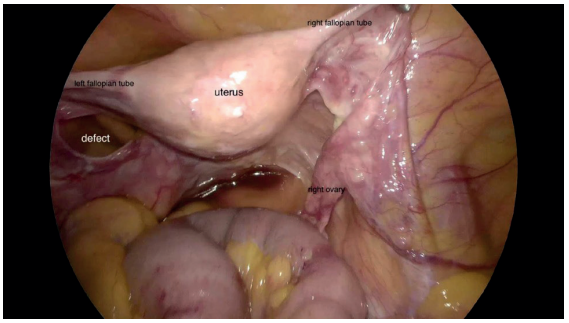

Figure 3. Appearance of the defect and anatomical landmarks after the ileal loops were reduced

two after confirming the absence of nasogastric drainage and disappearance of air-fluid levels on imaging. Oral intake was gradually increased, and the patient was discharged on postoperative day four.

Diagnosis: Type 1 Allen-Masters syndrome causing intestinal obstruction.

Informed Consent: An informed consent form was signed by the patient.

Discussion

Following Quain's initial report of herniation caused by a defect in the broad ligament, this anatomical anomaly has been further studied, with its etiology classified into two categories: congenital and acquired. The primary congenital cause is thought to involve the rupture of cystic remnants of embryological Mullerian ducts (10). Acquired causes, on the other hand, include iatrogenic injuries, pelvic inflammatory disease, or traumatic lesions. A broad ligament defect occurring after tearing during childbirth is referred to as Allen-Masters syndrome, which is commonly associated with chronic pelvic pain.

Defects in the broad ligament are classified based on their anatomical location or whether they involve the full thickness of the peritoneum. Cilley categorized broad ligament defects into three types according to the site of the tear. Type 1, the most common, occurs between the fallopian tube and the round ligament. Type 2 arises between the fallopian tube and the ovary, while Type 3 is located between the round ligament and the uterus (11). Similarly, Hunt proposed a classification with two distinct types (12). In Type 1, only one layer of the double-layered broad ligament is involved, forming a confined space where herniated structures can become trapped. Type 2, the more common form, involves a complete herniation through the entire ligament.

Internal herniation of the small intestine is a rare cause of small bowel obstruction, accounting for only 0.2% to 1% of all cases (6). Among these, only 4% to 5% are attributed to defects in the broad ligament, making it an exceptionally rare condition. While the ileum is most frequently involved, herniation of the colon, ovary, fallopian tube, and ureter has also been reported (6,13). Multiparity is a predisposing factor in approximately 80% of cases (9). The defect is typically isolated, and although the exact cause remains unknown, nearly three-quarters of cases demonstrate a left-sided predominance.

In this study, we reviewed previously reported cases of internal herniation through broad ligament defects, alongside our own case presentation. Using PubMed, we conducted a comprehensive search of all articles published up to November 2024. The search utilized the following terms: "Broad ligament AND internal hernia," "Broad ligament AND intestinal obstruction," "Allen-Masters syndrome AND

internal hernia," and "Allen-Masters syndrome AND intestinal obstruction." This search yielded a total of 115 articles. Among these, 97 articles were written in English, 85 of which were case reports. The remaining 18 articles were published in languages other than English, predominantly French, Spanish, Italian, Danish, Chinese, Japanese, and Russian. It was noted that the herniated intra-abdominal organ was most commonly the small intestine. The details of all identified studies and the cases presented in this article are summarized in Figure 4.

Adhesions are the most common cause of small bowel obstruction, accounting for 55% to 80% of cases in developed countries due to intra-abdominal and pelvic adhesions (14,15). Approximately 80% of patients with adhesive small bowel obstruction have a history of prior abdominal surgery (14). In the case we present, the patient had undergone two previous cesarean sections, and the presence of dilated intestinal loops with a transition zone in the pelvic region initially suggested an adhesive band involving ileal loops in the pelvis. The definitive diagnosis was confirmed through diagnostic laparoscopy, and treatment was successfully performed using a laparoscopic approach during the same session. In our case, the peritoneal defect was closed with absorbable sutures, in accordance with most recommendations in the literature (16,17). However, some studies, particularly those describing Cilley Type 1 defects, have reported alternative approaches, such as cleave the defect to prevent reincarceration (18).

As seen in our case, laparoscopy should be considered the first-line approach in the management of bowel obstructions caused by internal hernias, as it is less invasive and associated with better postoperative outcomes.

Conclusion

In conclusion, although adhesions and abdominal wall hernias are commonly considered in patients with signs of mechanical bowel obstruction and a history of intra-abdominal surgery, it is important to consider rarer etiologies. This case highlights Allen-Masters syndrome as a rare cause and emphasizes the importance of timely diagnosis and management of symptomatic broad ligament defects.

Ethical approval

In this case, we have ensured that the patient's identity is fully anonymized, and no identifiable information has been disclosed. Additionally,

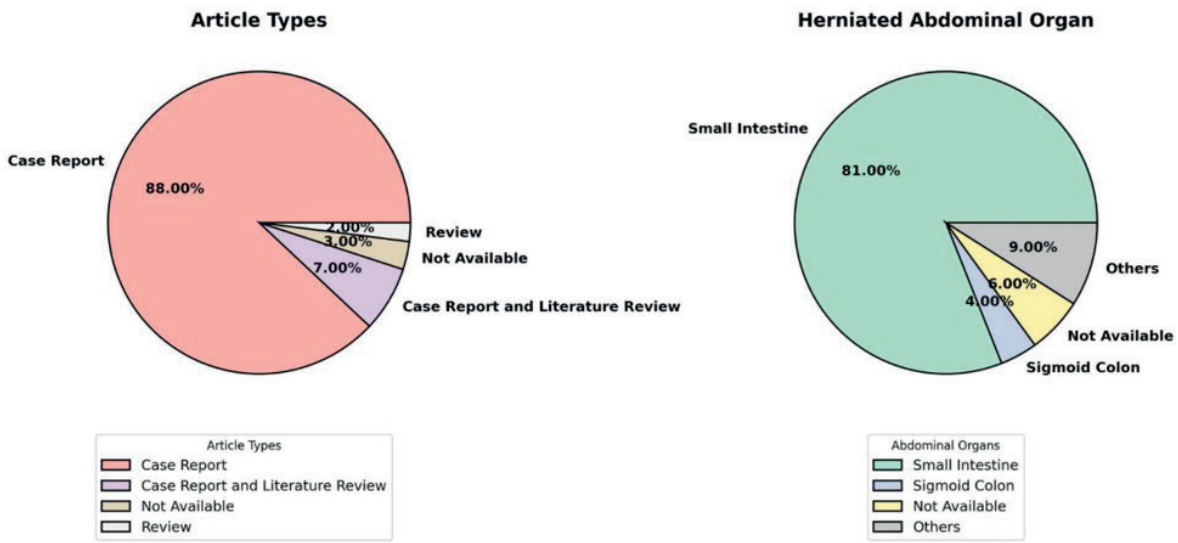

Figure 4. Characteristics of included studies

we have obtained the patient's informed consent to publish this report.

Author contribution

The authors confirm contribution to the paper as follows: Study conception and design: HOS, SY, YD; data collection: HOS, NA, OT; analysis and interpretation of results: HOS, SY, YD; draft manuscript preparation: HOS, NA, OT. All authors reviewed the results and approved the final version of the manuscript.

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