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Duodenal Intussusception Secondary to Hamartomatous Polyps of the Duodenum and a Review of the Literature

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1. Abstract

1.1. Introduction: Hamartomas of the duodenum are benign duodenal tumors comprising approximately 5-10% of duodenal tumors. The incidence is <0.01%. Typically asymptomatic, they may manifest as intestinal obstruction, gastrointestinal hemorrhage, biliary obstruction or intussusception. Intussusception as a manifestation of duodenal hamartoma is rare in itself with less than 200 cases reported in the literature.

1.2. Methods: A systemic literature review was performed encompassing all cases presenting with symptomatic intussusception secondary to a hamartamous polyp of the duodenum. Etiology of symptomatic intussusception included gastrointestinal hemorrhage, gastric outlet obstruction, gastrointestinal hemorrhage with concurrent gastric outlet obstruction and biliary obstruction. Treatments for each presenting etiology were explored.

1.3. Results: 17 cases of duodenal intussusception secondary to a hamartomatous polyp were identified. Two presented with gastrointestinal hemorrhage, seven with gastric outlet obstruction, five with a combination of the two and only three with biliary obstruction. Only one case was successfully treated with endoscopic polypectomy. Most were achieved by local resection (47%) followed by segmental bowel resection (24%). Two cases underwent reconstruction with a Billroth II approach and the other a Roux en Y gastrojejunostomy. Only one case underwent a pancreaticoduodenectomy.

1.4. Conclusion: Duodenal hamartomas are indeed a rare entity and typically asymptomatic. Even rarer are hamartomas resulting in symptomatic intussusception. Complications from these intussusceptions are far from few with only 17 described in the literature. Curative management can be achieved with less invasive methods such as local resection as almost half of presenting cases were treated this way avoiding more morbid measures such as gastrointestinal reconstruction and pancreaticoduodenectomy.

2. Introduction

Brunner's hamartoma, also known as Brunner gland adenoma or brunneroma, are benign duodenal tumors comprising approximately 5-10% of duodenal tumors [1-3]. The incidence of Brunner's hamartoma is <0.01% [4-6]. Typically asymptomatic, they may manifest as intestinal obstruction, gastrointestinal hemorrhage, biliary obstruction or intussusception [3,4,7,8]. Intussusception as a manifestation of a brunneroma is rare in itself with less than 200 cases reported in the literature.[9] There have only been 3 cases in the literature reported involving a Brunner's hamartoma causing intussusception with resultant biliary obstruction. We present the first case report of duodenal intussusception presenting with a partial mid-gut volvulus and biliary obstruction. In addition, a review of current literature and respective case series have been performed.

3. Case Report

24-year-old male with no significant past medical or surgical history, presented with 3 weeks of worsening epigastric abdominal pain and recently with weight loss, nausea, vomiting with oral intake and constipation for 3 days. His emesis was non bloody and non bilious in nature. He denied dark stools and his last bowel movement was 3 days ago. In the emergency department he was afebrile and hemodynamically stable. On physical exam he was softly tender to palpation in the epigastrium without distention. Laboratory work significant for a white blood cell count of 8.3, hemoglobin

13.2 with a hematocrit of 41.2, total bilirubin of 1.4 with a direct of 1.1, alkaline phosphatase 1109, AST/ALT of 85/206, lipase 411, amylase 284, creatinine 1.3 and potassium 2.5. He turned out to be COVID positive. Cross sectional imaging was performed in the form CT of the abdomen and pelvis with oral and IV contrast which revealed a very large intussusception seen in the right upper quadrant at the level of the duodenum with involvement of the ampulla. The intussusception contained multiple small bowel loops and mesenteric contents measured 7.5 cm transversely, 6.8 cm anterior-posteriorly and 12.8 cm cradiocaudal (Figure 1).
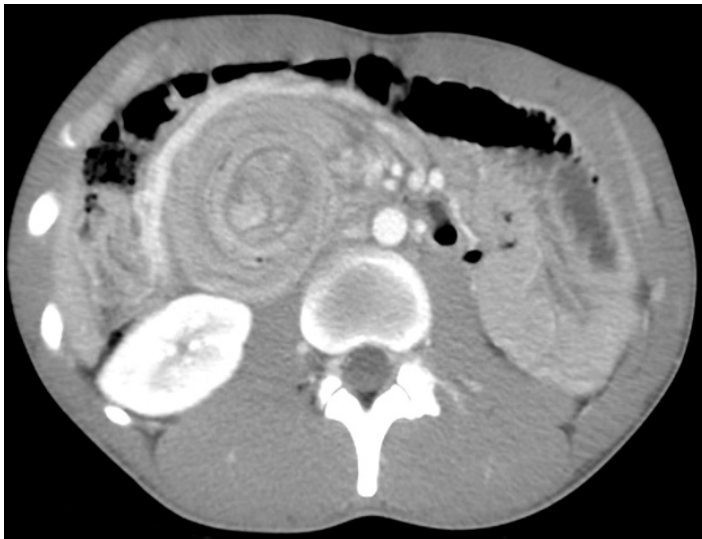

Figure 1: CT abdomen and pelvis representing intussusceptum containing multiple small bowel loops and mesenteric contents measured 7.5 cm transversely, 6.8 cm anterior-posteriorly and 12.8 cm cradiocaudal.

The stomach was collapsed. There was dilation of the main pancreatic duct, common bile duct and intrahepatic ductal dilation. The pancreatic duct measured 6 mm and the common bile duct 9 mm (Figure 2).
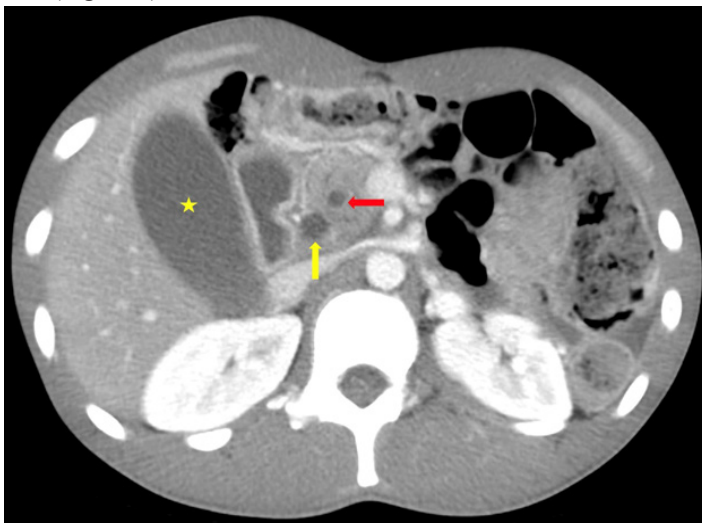
Figure 2: CT abdomen and pelvis representing dilated main pancreatic duct within intussusceptum (red arrow), dilated common bile duct (yellow arrow) and distended gallbladder (yellow star).

Given the findings on imaging, the patient was taken to the operating room for an exploratory laparotomy. Appropriate precautions were taken by all the OR personnel for COVID – 19 exposure. The duodenum was found to be significantly dilated and twisted around itself. Multiple loops of small bowel were intussuscepted into each other. The small bowel was detorsed initially to relieve the volvulus. The bowel seemed perfused well and there was active pulsations palpable in the small bowel mesentery. Next each intussusceptum was manually reduced from the mid jejunum upto the point of origin which happened to be the second portion of the duodenum. All of the bowel appeared viable. A duodenotomy was created at D1 just distal to the pylorus and was carried longitudinally along the antimesenteric border of the duodenum 5 cm distally. Two large intraluminal masses were found to be originating from the periampullary region pedunculated measuring approximately 10 and 12 cm in size. The ampulla did not seem directly involved. The decision was made to resect the masses at the base of the stalk without violating the ampulla. A common duct exploration was performed first. This began with a cholecystectomy in a top-down fashion. The cystic duct was accessed with a 4 French biliary Fogarty and the ampulla was intubated. This was used as a guide to protect the ampulla during the resection. Once the resection was complete at the level of the mucosa, the masses were sent for frozen section analysis to rule out an adenocarcinoma (Figure 3).
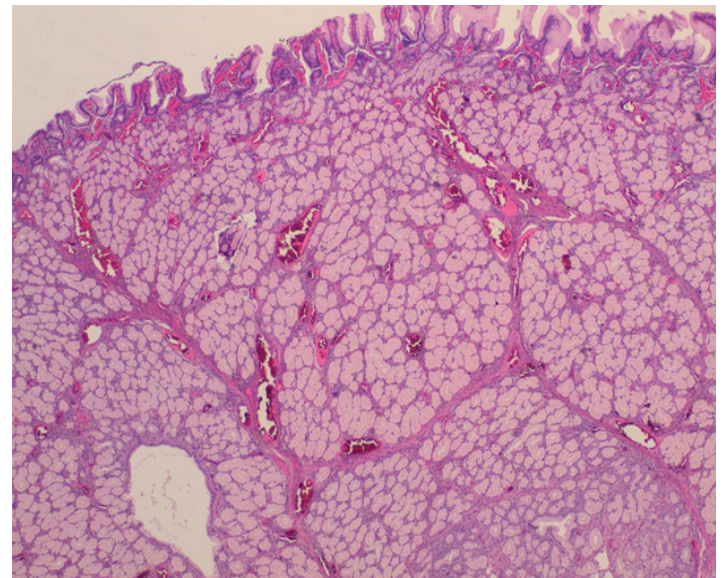
Figure 3: Gross Pathology: A tan-gray irregular, lobular/polypoid portion of tissue measuring 6.5 x 5.5 x 3.5 cm. On serial sectioning the specimen revealed tan-pink, lobular cut surfaces with multiple cysts measuring up to 0.4 cm in greatest dimension..

Figure 4: Microscopic Pathology: Large lobules of hyperplastic, benign Brunner's glands and numerous scattered dilated cystic spaces underneath a benign duodenal epithelium. The cystic spaces were lined by benign glandular, cuboidal and/or ciliated epithelium.

Preliminary diagnosis returned as duodenal hamartoma (Figure 4).

The mucosal edges at the site of resection were carefully approximated with 5-0 PDS suture. Finally, the anterior duodenotomy was primarily closed and an omental patch placed. The patient had a relatively normal postoperative course except for a prolonged ileus that eventually resolved with bowel rest and decompression. The patient being COVID positive did make the postoperative care challenging from a personnel standpoint.

4. Discussion

Intussusception is the prolapse of a proximal segment of bowel into a distal segment. It is most commonly seen in children and less commonly in adults [10]. Intussusception in adults account for only 5-16% of all intussusceptions [11]. Pathogenesis usually involves a lead point such as Meckels' diverticulum, hamartomas, hypertrophied Peyer's patches and tumors [10,12]. These masses in the bowel wall or lumen are thought to provoke abnormal peristalsis leading to the telescoping of bowel segments [13]. Due to the duodenum being mostly fixed in a retroperitoneal position, duodenal intussusception is relatively rare [11,14]. Duodenal intussusception can occur with involvement of the stomach, jejunum, or strictly duodenum, also known as duodeno-duodenal intussusception. Most duodenal intussusceptions are due to tumors, adenomas or Brunner's gland hamartomas [9]. Brunner glands are located primarily in the proximal duodenum and duodenal bulb that serve as a barrier from gastric acid by secreting alkaline fluid into the duodenum [3]. Abnormal growth of the glands is known as Brunner gland hyperplasia, also known as Brunner's hamartoma, brunneroma or Brunner's gland adenoma and subclassified into three categories: diffuse hyperplasia, circumscribed hyperplasia and glandular adenoma [3]. The pathogenesis of hyperplasia of the Brunner gland is unknown however theories include chronic renal failure, chronic pancreatitis, peptic ulcer disease or H. pylori infection [1,15,16]. Brunneromas comprise 5-10% of all benign duodenal tumors [1,2,3,17,18]. The duodenal bulb is the most common location of a Brunner gland hamartoma accounting for 70% [17]. The second most common location is the second portion of the duodenum accounting for 26% of all brunneromas [8,13]. The reason for this is Brunner's gland are in their highest concentration beginning at the duodenal bulb and decrease in number distally [1]. Most hamartomas are found incidentally on upper gastrointestinal series or endoscopy as they are typically asymptomatic [2,3,15,19]. Although exceedingly rare, symptoms appear once the tumor is > 2 cm [8,20,21,30]. The most common presenting symptoms are non-specific including abdominal pain, nausea, bloating and melena [1,22]. The most common presenting complications of duodenal intussusception are gastrointestinal hemorrhage and obstruction which are quoted from 37-45% and 37-50% respectively in the literature [1,3,9]. Typical imaging studies in the workup include barium swallow, ultrasound, CT, endoscopic ultrasound and upper endoscopy [1,2,9,10,16,23]. Barium swallow reveals a mobile, pedunculated polypoid intraluminal filling defect with smooth borders and the classic "coiled spring" [2,22,24]. Filling defects are non-specific however. Findings may mimic other duodenal tumors such as leiomyoma, lymphoma or lipomas [1,25]. Endoscopic biopsies are usually negative and only reveal hyperplasia of the Brunner's gland due to the submucosal location of the mass [1,17]. Endoscopic ultrasound may be useful to prove that there is no involvement of the muscularis mucosa which argues for a diagnosis of Brunner's harmatoma [1,9,26]. CT is the typical choice of imaging as it reveals the size of the mass, internal characteristics, location, relationship to other structures, intussusception and obstruction. The draw back as with endoscopy is the inability to distinguish a Brunner's hamartoma from other duodenal tumors [17]. There is still much debate whether small, incidentally found, hamartomas require removal. Reports of malignant foci have been described in the literature and therefore resection whether endoscopic or surgically is recommended [3,10]. Other reasons include confirmation of suspected diagnosis, rule out malignancy and to prevent or treat complications such as gastrointestinal hemorrhage and obstruction [1,37]. Management includes endoscopic polypectomy and open versus laparoscopic resection. Endoscopic polypectomy is the treatment of choice for small tumors [2]. Surgical resection includes local excision through a duodenotomy, segmental resection of bowel, pancreas-sparing duodenectomy or pancreaticoduodenectomy [17]. In the review of the literature, there were only 17 cases of duodenal intussusception secondary to a Brunner's gland hamartoma. Two presented with signs of gastrointestinal hemorrhage, seven with gastric outlet obstruction, five with a combination of the two and only three with biliary obstruction (Table 1). Furthermore only one case was successfully treated with endoscopic polypectomy. Most were achieved by lo-

cal resection (47%) followed by segmental bowel resection (24%). Two cases underwent reconstruction with a Billroth II approach and the other a Roux en Y gastrojejunostomy. Only one case had to undergo a pancreaticoduodenectomy secondary to invasion of the pancreatic head [4]. The three cases of the rarest presentation, biliary obstruction, were reported in 1995, 2016 and 2019. In 2016 resection was achieved by local excision and In 2019 by segmental bowel resection. Unfortunately, management was not described in the report of biliary obstruction in 1995 [5,11,28]. From a paper in 2019 quoted the largest Brunner's gland hamartoma to be 12 cm [5,29]. On our review we found only two incidents of Brunner's gland hamartomas causing duodenal intussusception to be 12 cm in size from 1991 and 2019 [5,22].

Patient Presentation and Intervention Performed

	Endoscopic Polypectomy	Local Excision	Bowel Resection	Whipple	R-Y	Unknown	Symptom Frequency
GIB	0	1	1	0	0	0	2 (12%)
GOO	0	4	1	1	1	0	7 (41%)
GIB + GOO	1	1	2	0	1	0	5 (29%)
BO	0	2	0	0	0	1	3 (18%)
Intervention Frequency	1 (6%)	8 (47%)	4 (24%)	1 (6%)	2 (12%)	1 (6%)	17

Table 1: Patient presentations are divided into gastrointestinal bleed (GIB), gastric outlet obstruction (GOO), gastrointestinal bleed with gastric outlet obstruction (GIB + GOO) and biliary obstruction (BO). Interventions for treatment performed include endoscopic polypectomy, local excision, bowel resection, Whipple and resection with Roux-en-Y reconstruction (R-Y). There are a total of 17 cases and 16 interventions reported.

5. Conclusion

Brunner's hamartoma are indeed a rare entity and typically asymptomatic. Even rarer is duodenal Intussusception secondary to a Brunner's hamartoma. Complications from these intussusceptions are far from few with only 17 described in the literature. Complications include gastrointestinal hemorrhage, gastrointestinal obstruction and biliary obstruction. Imaging of choice is CT of the abdomen and pelvis. Definitive diagnosis and treatment is made by excision and pathologic review. Excision is obtained from least to most invasive: endoscopic polypectomy, local excision, segmental bowel resection with or without reconstruction and pancreaticoduodenectomy. From the literature reviewed, our case appears to be one of the most unique with rare traits. Volvulus along with intussusception has never been described in the literature. We are the fourth to present a case of intussusception with biliary obstruction. Lastly, ours was one of if not the largest Brunner's gland hamartoma with two hamartomas 12 cm in size each on resection.

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