ISSN: 2755-015X

Journal of Surgery & Anesthesia Research

SCIENTIFIC Research and Community

Case Report

Open Access

Small Bowel Adenomyoma: An Incidental Intraoperative Finding While Performing Hepaticojejunostomy for CBD Injury

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ABSTRACT

Adenomyoma is a benign lesion that is most commonly seen in the gallbladder, however, rare cases have been reported where this pathology was encountered in the vicinity of the gastrointestinal tract. The pathogenesis of this lesion is still a controversy, with the previous reports suggesting it to be either a form of hamartoma or incomplete heterotopic pancreas. Jejunal and ileal adenomyoma have been rarely reported, and as of 2016 less than 30 cases were reported in the English literature. The clinical presentation is variable depending on the location of the lesion. Although there are no specific management guidelines for this pathology, a surgical resection is sufficient. However, aggressive surgical approaches, such as pancreaticoduodenectomy for periampullary adenomyoma, have been undertaken in the previous reports due to the misdiagnosis with carcinoma preoperatively. We report a case of a 58-year-old gentleman who was referred to our Hepato- Pancreato-Biliary facility with common bile duct injury post laparoscopic cholecystectomy for hepatico- jejunostomy. Intra-operatively, an intra-luminal, jejunal mass was found measuring 2x2 cm and was about 95 cm from the DJ junction. The lesion was resected with safety margins, and primary anastomosis was done. The final histopathology of the specimen was consistent with adenomyoma, and all of the surgical margins were free.

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Received: September 10, 2020; **Accepted:** September 17, 2020; **Published:** September 19, 2020

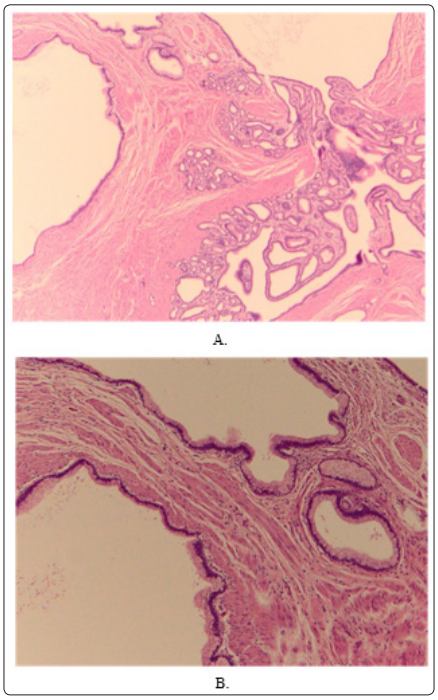
Keywords: Adenomyoma, Small Intestine, Jejunum, Case Report

Introduction

Adenomyoma is a benign lesion that's most commonly seen in the gallbladder [1]. However, rare cases have been reported where this pathology was encountered in the vicinity of the gastrointestinal tract; most frequently in the antrum of the stomach, followed by the periampullary area of the small intestine, and less than 40 reported cases occurred in the jejunum and the ileum. The pathogenesis of this lesion is still a controversy, with the previous reports suggesting it to be either a form of hamartoma or incomplete heterotopic pancreas [1,2]. We present a case of an incidental jejunal adenomyoma found intraoperatively and managed with a segmental resection. The work has been reported in line with the SCARE criteria [3].

Case Report

A 58-year-old gentleman, with a long-standing history of hypertension, was referred to our HPB facility with a common bile duct (CBD) injury, due to proximal ligation during laparoscopic cholecystectomy as seen on initial imaging and manifested 2 days postoperatively with persistent pain, and obstructive jaundice. The patient was admitted at our institute 3 months after his operation for definitive surgical management. During that period a percutaneous trans-hepatic biliary drainage (PTD) tube, for external drainage, was inserted to relieve his symptoms. Physical examination was otherwise unremarkable except for mild jaundice. His initial laboratory findings were within acceptable range apart from a deranged hepatic function as follows: total bilirubin 43(Normal Range 4- 21umol/L), direct bilirubin 35.6(normal range 0.8-3.3 umol/L), alkaline- phosphatase 414(normal range 54-144 u/L), Alanine Aminotransferase 213(normal range 17-45 u/L), Aspartate Aminotransferase 105(normal range 8- 38 u/L). Computed tomography (CT) scan and Magnetic resonance cholangiopancreatography (MRCP) revealed prominent intrahepatic biliary radicals with PTD in place. A cystic lesion measuring 5 cm was noted along the lesser curvature and occupying the gastrohepatic ligament and the lesser sac, however, it was not communicating with the pancreaticobiliary tree likely representing a mesenteric cyst or lymphocele. A duodenal diverticulum was also noted along the posteromedial wall of the 2nd part. No lesion was detected along the bowel wall. The patient underwent an exploratory laparotomy with Roux- Y hepaticojejunostomy. During the formation of the R-Y jejunal limb, an intraluminal firm mass was felt in the jejunum measuring 2 cm in size and was 95 cm away from the DJ junction. A limited segment of the jejunum containing the mass was resected with distal and proximal safety margins and side-side anastomosis was done. Final histopathology review revealed a 1x1x0.8 cm, whitish fungating mass, with a haphazard proliferation of bland, cystically dilated, and mucin secreting glands, surrounded by smooth muscle fibers; features consisting with adenomyoma (Figure 1). The patient had an eventful recovery and was discharged home in a stable condition on the 7th post-operative day.



Figures 1: Histological picture: Hematoxylin and eosin–stained section of the mass showing haphazard proliferation of bland, cystically dilated, mucin secreting glands surrounded by smooth muscle fibers [A: Low power, B: High power]

Discussion

Adenomyoma, or also known as myoepithelial hamartoma, adenomatous hamartoma or foregut choristoma, is a benign tumor-like lesion frequently encountered in the gallbladder [1-4]. The gastrointestinal tract is the second most frequently reported site for this lesion, although rare, with the majority of the cases occurring in the antrum of the stomach and the periampullary area of the small bowel, where it may present with biliary obstruction and can be misdiagnosed as a malignant lesion requiring extensive surgical resection [1,2,5]. Jejunal and ileal adenomyoma have also been reported to a lesser extent in the literature, and as of 2016 less than 30 cases were reported in the English literature. This low incidence can be explained by underreporting this lesion when discovered, as they can represent an incidental finding at autopsy. Jejunal lesions are by far the least frequent of all small bowel adenomyoma and comprise less than a third of the cases [2,4,6].

Adenomyoma is often described as duct-like, glandular structures, lined by cuboidal-columnar epithelium and surrounded by interlacing bundles of smooth muscle [2,7]. Occasionally, the lesion may also contain Goblet cells, the glands might show cystic changes. Moreover, this lesion often tends to be located in the submucosa and may extend to the muscularis propria [2,4,8,7,9].

The origin of gastrointestinal adenomyoma is still unclear, and despite having similar histologic components, it's pathogenically different from gallbladder adenomyoma that arises from a diverticular disease of the gallbladder [1,10]. The two widely accepted theories explaining the pathogenesis are the intestinal hamartoma and incomplete pancreatic heterotopia theories [1,2,7,11]. A hamartoma refers to the presence of excessive, however, focal growth of tissue elements that are native to the organ where they are; and since adenomyoma presents a nodular lesion with all intestinal tissues, alongside with the finding of transitional area between the epithelial component of adenomyoma and the overlying intestinal mucosa, the hamartoma hypothesis was a valid theory for some authors [2,12]. However, the fact that the lesion also contains duct-like glandular structures that stain positive to CK7 and CA 19-9, expressed by the pancreatic ductal epithelium, and stain negative for CK20 and CDX2, expressed by the intestinal epithelial cells, makes the pancreatic heterotopia theory more prevailing and widely accepted by most of the authors; however, the lesion actually lacks the presence of pancreatic acinar and islet cells, thus making it an incomplete form of pancreatic heterotopia [1,2,13,7,12]. Heterotopia (Choristoma) refers to the growth of normal cells in an anatomically abnormal place, and likewise, pancreatic heterotopia is defined as normal pancreatic elements without anatomic or vascular connection to the gland [12, 14,15]. There are two theories that explain this phenomenon, the migration theory, in which fragments of the normally developing pancreas detach during the foregut rotation to rest in an aberrant tissue; and the metaplasia theory where the totipotent cells, of endodermal origin, give rise to pancreatic tissue in an anomalousplace [14]. Pancreatic heterotopia is classified into 3 categories based on the extent of differentiation of the pancreatic elements. In type I the tissue has ducts, acini and islet cells; in type II the ducts are present along with the acini only, and type III is devoid of acini and islet cells with the mere presence of ducts; this type is what's agreed on to be called adenomyoma [15]. The presence of smooth muscle cells can be explained by secondary proliferation caused by some sort of stimulus from the aberrant epithelium, whereas the presence of Goblet cells and other intestinal elements could result from metaplasia [1,2].

The clinical presentation of patients with small bowel adenomyoma is variable based on the location as well as the patient's age; however, many cases were reported to be incidentally found upon autopsy or intraoperatively for other causes. As far as the ileal and jejunal lesions are concerned, the most frequently reported complication is intussusception, however, bowel obstruction and lower gastrointestinal bleeding (LGIB) have also been reported [5,13,9]. Adenomyoma and myoepithelial hamartoma are not known to be associated with genetic, congenital disorders or familial syndromes, however, at least 3 cases were reported with such rare association. The first case was reported by Hizawa et al where a jejunal adenomyoma was found incidentally as a submucosal tumor among other adenomas upon evaluating a patient with a known family history of Gardner's syndrome [16]. Another rare finding was the case reported by Yao et al in the year 2000, where an adenomyoma was found in Meckel's diverticulum causing intussusception in a 22 month-old boy. This case, in particular, gives as a strong-evidence that adenomyoma is, in fact, a form of pancreatic heterotopia. Lastly, Koh et al reported a case where jejunal adenomyoma was found incidentally in a 27-year-old lady who's known to have congenital bowel malrotation. Based on the current literature, the median age at diagnosis was 25 years (ranging from 2 days to 82 years), with a bimodal distribution of the cases having 2 peaks; < 30 years (where the majority is) and > 50 years of age [2,8]. In the younger age group, there was a male predominance and the lesion was found more frequently in the ileum than the jejunum causing intussusception or small bowel obstruction. On the other hand, in the elderly age group, the lesion was more or less found incidentally during surgery for other pathology [8]. Table 1 shows the clinicopathological characteristics of ileal and jejunal adenomyoma reported in the cases from 1940 till 2018; 26 of which were already reviewed in previous papers, and we identified 11 more cases including ours added to the list.

Summary of reported cases of ileal and jejunal adenomyoma (Clinico-pathological characteristics)

Serial no. of cases in each series	Author and year of publication	Age	Sex	Presentation	Location	Management
1	Clarke et al. (1940)	64 years	Male	Incidental finding	Jejunum	N/A
2	Schwartz et al. (1958)	8 months	Male	Intussusception	Ileum	Segmental resection
3	Benisch et al. (1978)	47 years	Female	Incidental finding	Ileum	Segmental resection
4	Rosenmann et al. (1980)	2 days	Female	Intestinal obstruction	Ileum	N/A
5	Gal et al. (1986)	82 years	Female	Intussusception	Ileum	Segmental resection
6	Kim et al. (1990)	7 years	Male	Intussusception	Ileum	N/A
7	Gal et al. (1991)	9 months	Male	Intussusception	Ileum	Segmental resection
8	Gal et al. (1991)	79 years	Male	Incidental finding	Ileum	Segmental resection
9	Lamki et al. (1993)	1 year	Male	Intussusception	Ileum	N/A
10	Serour et al. (1994)	3 years	Male	Intussusception	Ileum	N/A
11	Chan et al. (1994)	5 months	Female	Intussusception	Ileum	Segmental resection
12	Chan et al. (1994)	3 years	Male	Incidental finding	Ileum	Two Segmental resection (involving the jejunum and ileum)
13	Gonzalvez et al. (1995)	Gonzalvez et al. (1995)	Male	Intussusception	Ileum	Segmental resection
14	Tanka et al. (1996)	24 years	Male	LGIB	Ileum	Segmental resection
15	Hizwa et al. (1996)	23 years	Female	Incidental finding	Jejunum	Segmental resection
16	Yamagami et al. (1997)	4 months	Male	Intussusception	Ileum	Wedge resection
17	Van Helden et al. (1998)	64 years	Male	LGIB	Jejunum	Segmental resection
18	Yao et al. (2000)	1 year	Male	Intussusception	Meckel's diverticulum	Resection of Meckel's diverticulum
19	Ueyama et al. (2001)	52 years	Female	Incidental finding	Ileum	Wedge resection
20	Lee et al. (2002)	18 years	Male	Intussusception	Jejunum	Segmental resection
21	Mouravas et al. (2003)	1 year	Male	Intussusception	Ileum	Segmental resection
22	Gemma et al. (2003)	13 years	Female	Intussusception and volvolus	Jejunum	N/A
23	Park et al. (2003)	7 months	Male	Intussusception	Ileum	Segmental resection
24	Park et al. (2003)	63 years	Male	Incidental finding	Jejunum	Left hemicolectomy and jejunal segmental resection
25	Koh et al. (2003)	27 years	Female	Incidental finding; known to have bowel malrotation;	Jejunum	Segmental resection
26	Takahashi et al. (2006)	75 years	Male	Incidental finding	Ileum	None; autopsy finding
27	Ikegami et al. (2006)	5 months	Female	Intussusception	Ileum	Segmental resection including the jejunum
28	Yu et al. (2008)	74 years	Female	LGIB	Jejunum	Segmental resection
29	Qing et al. (2009)	61 years	Female	Incidental finding	Jejunum	Segmental resection

30	Cansiz et al. (2010)	61 years	Male	Strangulated inguinal hernia	Jejunum	Segmental resection and primary hernia repair
31	Takeda et al. (2011)	68 years	Male	Intussusception	Ileum	Segmental resection
32	Tomibayahshi et al. (2011)	81 years	Female	Intestinal obstruction	Jejunum	Segmental resection
33	Bak et al. (2014)	11 months	Female	Intussusception	Ileum	Segmental resection
34	Khmou M.et al.(2015)	29 years	Male	Intussusception	Ileum	Segmental resection
35	Hajimaghsoudi et al. (2016)	9 years	Male	Incidental finding	Ileum	Appendectomy and ileal resection
36	Pitto et al. (2016)	86 years	Female	LGIB	Jejunum	Segmental resection
37	Present case (2018)	58 years	Male	Incidental finding	Jejunum	Segmental resection
NA: not available, LGIB: Lower gastrointestinal bleeding						

In general, the nature of the lesion is benign and is not part of a neoplastic process on the premise of lacking atypia on histopathologic examination and the low proliferative activity on immunohistochemical staining; yet, periampullary lesions can be misdiagnosed preoperatively with malignant tumors due to the similar clinical presentation [1,5,7].

Thus, some authors advocate for intraoperative frozen section to avoid aggressive surgery for such a benign tumor.These lesions, however, don't require more than a simple surgical resection[1,2]. Table 1 also shows that most of the authors elected to resect the tumor, and segmental resection was enough in all cases[17].

Conclusion

Small bowel adenomyoma is a rare, benign lesion that can be encountered incidentally during laparotomy for other causes. Being aware of the benign nature of this lesion help avoid unnecessary extensive surgical management as simple resection usually suffices.

Disclosure

The authors declare that there is no conflict of interest regarding the publication of this paper.

Acknowledgment

This work has been presented in Abstracts of the Americas Hepato-Pancreato Biliary 2019 Annual Meeting.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Contribution

HO wrote the manuscript and reviewed the literature. AM drafted the manuscript and reviewed the literature. MT reviewed the final manuscript. MQ supervised the management of the patient and reviewed the manuscript. All authors read and approved the final manuscript.

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