

## Adenomyoma of the Small Intestine in a Child, A Case Report

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<http://dx.doi.org/10.13005/bbra/2305>

(Received: 20 July 2016; accepted: 03 September 2016)

**Adenomyoma is a tumor including components derived from glands and muscle. The main position of Adeomyoma occurrence is the stomach and the second most frequent location is the small bowel, usually in the periampullary area. and its occurrence in the small intestine distal to the duodenum is very rare. A 9 years old male patients admitted to the hospital with abdominal pain, nausea, and vomiting. The patient underwent surgery with acute appendicitis diagnosis. In addition to the inflammation of the appendix, a 3 cm nodule within 150 cm of ileocecal valve without any adhesion to the surrounding tissues was observed during the surgery. Our histopathological findings of this patients did not show goblet cells in all part of lesion. Adenomyom of ileum is rare disorder and the location of this lesion was not reported before.**

**Keywords:** Adenomyoma, Goblet cells, Ileum, Child.

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Adenomyoma is a tumor including components derived from glands and muscle<sup>1</sup> It is a type of complex and mixed tumor. In obstetrics and gynecology contexts, it is a form of adenomyosis that forms a mass or growth around the tissue of the inner uterus. Most cases of adenomyosis are non-symptomatic. However, it may present with dysmenorrhea and pelvic pain. In case of juvenile cystic adenomyoma, laparoscopic enucleation results in a statistically and clinically significant reduction in dysmenorrhea, ease in any chronic pelvic pain and low risk of recurrence<sup>2</sup>

Adenomyoma of the gastrointestinal tract is a benign tumor-like condition characterized

histologically by abnormal glandular formations lined by mucin-secreting columnar epithelium and surrounded by bundles of smooth muscles<sup>3-5</sup> Magnus Alsleben, in 1903 was the first one who described adenomyomas with stomach localization<sup>6</sup> It also has several other names such as myoepithelial hamartoma<sup>7-11</sup>, adenomyomatous hamartoma<sup>12,13</sup>, and foregut choristoma<sup>14</sup>. It arises from pancreatic metaplasia or a displaced pancreatic anlage during embryogenesis<sup>15</sup>

The main position of Adeomyoma occurrence is the stomach<sup>1</sup> and the second most frequent location is the small bowel, usually in the periampullary area<sup>16</sup> and its occurrence in the small intestine distal to the duodenum is very rare<sup>17</sup> The most common clinical presentations of adenomyoma in the small bowel are intussusception and intestinal or biliary obstruction, which is depend on the location of

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the lesions<sup>5, 7, 12, 13, 18</sup> Although most cases refer to adults, they may also appear at any age<sup>19</sup> and it also has been seen in children<sup>20</sup> Herein we try report a 9 years old patient who suffer from adenomyoma in the small intestine distal and near to ileocecal valve.

#### Case presentation

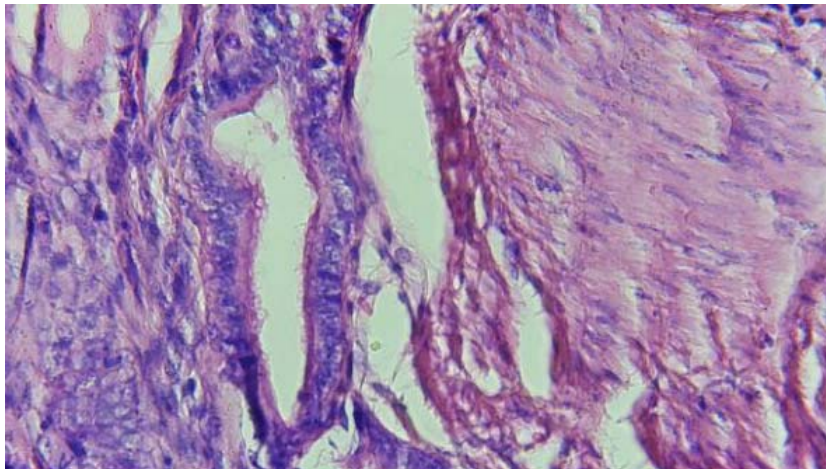
A 9 years old male patients admitted to the Bahonar hospital, Karaj with abdominal pain, nausea, and vomiting. Due to the investigation, the patient did not have any history of a particular disease in past. Lower abdominal pain and tenderness with leukocytosis and urine turbidity, and small renal calculus in the lower calyx of left kidney and lymph node with diameter up to 0.7 mm were our other findings obtained from this patient.

The patient underwent surgery with acute appendicitis diagnosis. In addition to the inflammation of the appendix, a 3 cm nodule within 150 cm of ileocecal valve without any adhesion to the surrounding tissues was observed during the surgery. Histopathologic examination showed that the nodule lesion made up of pancreatic cystic ducts with smooth muscle tissue. Pancreatic glandular components were not observed in this tissue. These pathological findings consistent with adenomyoma in this area. (Figure 1 and 2)

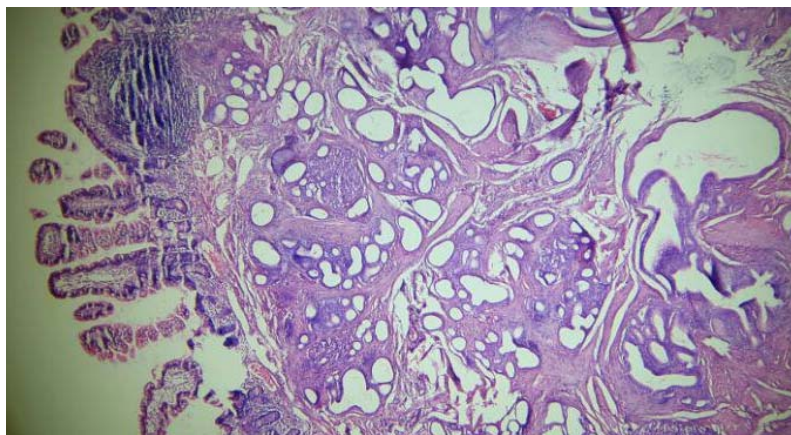
The young patient discharged after the surgery and standard diet with a normal postoperative course.

#### Discussion

Adenomyoma of the gastrointestinal (GI) tract is a rare lesion and only a small number of



**Fig. 1.** Histological image. Adenomyoma of jejunum, dilated exocrine type cyst. high power ( $\times 400$ ), H&E stained



**Fig. 2.** Histological image. Adenomyoma of jejunum, smooth muscle bundles between dilated and cystic exocrine type cysts. low power ( $\times 100$ ), H&E stained

**Table 1.** Summary of reported adenomyoma located in ileum

Study	Age/Sex	Size (cm)	Location
Roseman et al [22]	2 Days/F	NA	Ileum, submucosa
Kim et al [14]	7 Years/M	4	Ileum, submucosa
Gal et al [12]	9 Months/M	1.2	Ileum, submucosa, and muscularis
Serour et al [24]	4 Years/M	2	Ileum, Submucosa
Chan and Roche [3]	5 Months/F	0.8	Ileum, Submucosa
Schwartz et al [25]	8 Months/M	2	Ileum
Lamaki et al [26]	1 Year/M	1	Ileum
Gonzalvez et al [8]	2 Years/M	2	Ileum
Yamagami et al [11]	4 Months/M	NA	Ileum
Mouravas et al [19]	1.5 Year/M	1	Ileum
Park et al [13]	7 Months/M	1.2	Ileum
Temiz et al [27]	11 Years/M	1.5	Ileum
Bak et al [28]	11 Months/F	1	Ileum
Current Report	9 Years/M	3	Ileum

NA: Not Available

them occur in the small bowel distal to the duodenum<sup>12</sup> Symptoms of adenomyoma of the GI tract depend on the location of the lesion and patient's age. Adenomyoma of the periampullary area usually presents with biliary obstruction (obstructive jaundice) or abdominal pain<sup>21</sup> Jejunal and ileal adenomyoma of pediatric patients usually presents with intussusceptions<sup>22</sup> In adult patients, intussusception is an infrequent complication, with many reported cases having been incidentally detected during surgery for other diseases or during autopsy. Several cases presenting with GI bleeding (melena) have been reported<sup>10, 23</sup>

The pathogenesis of adenomyoma of the GI tract is generally considered to be either a form of hamartoma or a pancreatic heterotopia, although it is not fully understood. The term 'hamartoma' means an excessive but focal overgrowth of cells or tissues native to the organ in which it occurs. The term 'heterotopia' means microscopically normal cells or tissues existing in an abnormal location. The existence of goblet and Paneth cells in the glandular element of the present case seemed to suggest that the lesion was a true hamartoma<sup>17</sup>

Their localization at the small intestine of children is extremely rare (Table 1). Intussusception is the most common complication of adenomyoma of the small intestine, as was the case with our

patient. Thus far, of the 13 reported cases of adenomyoma in children, only two did not cause intussusception: in one case, intestinal obstruction preceded, and the other concerned an incidental finding<sup>3, 19, 22</sup> It is notable that this disorder also has been seen in higher ages and not shown here. These reports shows that adenomyoma do not have any relation to the patients age. Based on comparison of other reports it can be seen that this lesion at ileum is more common in male children.

Adenomyoma of the small intestine is formed by glandular structure lined by columnar epithelium and surrounded by smooth muscle. Cystic changes often occur in the ducts. Adenomyoma and ectopic pancreatic tissue can be seen in a variety of pathological changes. Acinar and pancreatic islet tissue usually can be seen in ectopic pancreatic tissue<sup>13</sup> Sever gastrointestinal bleeding is rare symptom of both these disorders. These nodules are often a random finding during abdominal surgery. Anatomically, adenomyom location at distal duodenum is very rare. Only 26 percent of all reported adenomyoms have been located at small intestine. These nodules can be manifested as an intussusception in children. The size of nodules can be varying from 0.6 to 6 cm<sup>15</sup>

Based on pathological studies, the goblet

cells observed only in 5 of 22 patients with small intestine adenomyom. <sup>17</sup> Our histopathological findings of this patients did not show goblet cells in all part of lesion.

Abdominal pain can be mentioned as a common presenting symptom in these patients. Hamartoma polyps in peutz jegher syndrome and/or metastatic adenocarcinoma can be mentioned as pathological differential diagnosis of this disorder. The absence of Desmoplastic stroma and also lack of cellular atypia are two important criteria for differentiation of malignant cases<sup>18,23</sup>

In summary, adenomyom of ileum is rare disorder and the location of this lesion was not reported before. On the other hand, goblet cells were not separated in all part of the nodule base on histopathological findings.

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