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Ultrasound diagnosis of the small intestinal adenomyoma in children

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Abstract

Aim: Adenomyoma is an exceptionally rare hamartoma in the small intestine. Few data have been reported on the features of this rare disease. The aim of this study was to describe the ultrasound (US) characteristics of small intestinal adenomyomas. **Material and methods:** This retrospective study analyzed the clinical features and US data of 15 pediatric patients diagnosed as small intestinal adenomyomas in the age range between 1 day to 12 years in our hospital during 2014–2021. **Results:** The clinical manifestations of all the small intestinal adenomyomas were abdominal pain, vomiting or/and hemafecia. The small intestinal adenomyoma usually acted as the lead point of secondary intussusception. They were identified in the ileum (n=11), jejunum (n=2), and Meckel's diverticulum (n=2). The diagnostic accuracy (the concordance rate between US diagnosis and pathological diagnosis) of small intestinal adenomyoma was 73.3%. The small intestinal adenomyoma had approximately 1.0–3.0 cm, were typically located in the submucosal region, had the basal part wide and without a pedicle, and its boundaries were clear. The mass protruded into the intestinal cavity, and showed oval hypoechoic polycystic echo nodules, containing multiple small quasi-circular or irregular cysts of different sizes surrounded by solid hypoechoic mosaic areas. The color Doppler US showed in the solid hypoechoic areas of the mass abundant or sparse blood flow signals. **Conclusions** The US findings of small intestinal adenomyomas in children are characteristic, and US is valuable in the identification of intestinal adenomyomas in children.

Keywords: intestinal adenomyoma; ultrasound; intussusception; children

Introduction

Adenomyoma, a rare benign tumor of the gastrointestinal system [1,2] also known as foregut choristoma, hamartoma, myoepithelial, or adenomyomatous [3-5] was first described by Magnus-Alsleben in 1903 [6]. In 1940, Clarke suggested that adenomyoma was induced by variable differentiation of abnormal embryonic epithelial buds [7]. Although adenomyoma was first classified as a hamartoma in 1991, it is generally thought to be a form of epithelial hamartoma or incomplete heterotopic pancreas [1,8-10]. Histologically, adenomyomas are characterized by glandular structures lined with a layer of epithelium that is cuboidal to tall columnar in shape and surrounded by bundles of smooth muscle cells. However, the pathogenesis of adenomyomas remain unclear [11].

Adenomyomas usually occur in the stomach, duodenum, and periampullary area, but rarely can occur also in the jejunum and ileum [2-4]. The occurrence of adenomyomas in the small intestine is exceptionally rare in children, with only 27 cases described in 23 case reports [1-4,7,8,12-31]. Adenomyomas in the periampullary area usually present as biliary obstruction or abdominal pain, whereas jejunal and ileal adenomyomas usually cause intussusception and present as abdominal pain [12]. Almost all cases of adenomyoma were diagnosed during surgical procedures for intussusception or autopsies. Endoscopic or radiological examination yields uncharacteristic findings, and there is no description of ultrasound

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(US) aspect of small intestinal adenomyomas, except for a single case reported by our team [28].

This study retrospectively reviewed the clinical features and US characteristics of pediatric cases of adenomyoma confirmed by surgery and pathology at our hospital. It is important for clinicians and sonographers to maintain their current knowledge of this disease and avoid making inaccurate diagnosis. The aim of this study was to describe the US features of small intestinal adenomyomas.

Material and methods

Our retrospective analysis included medical records of pediatric patients with intestinal adenomyoma who were diagnosed and underwent operations in our hospital between 2014–2021. We searched surgical patient data using the following keywords: intestinal adenomyoma, intussusception, Meckel's diverticulum, or cystic mass. Fifteen patients pathologically diagnosed with intestinal adenomyomas were enrolled in this study. For each patient, data on demographic characteristics, clinical presentation, US images, treatment, and pathological results were collected.

All the patients were scanned by an experienced pediatric sonographer with linear array (12-5 MHz) transducers of IU Elite, IU EPIQ, or IU22 units (Philips Medical Systems, Bothell, WA). The routine abdominal US was carried out according to the standard of US examination in our hospital, including gray scale with a gradual increase in pressure and color Doppler US, multi-section and multi-angle observation, and the relevant images were archived. Two experienced pediatric sonographers (more than 10 years) independently reviewed US images on a picture archiving and communication system. If there was disagreement between the results, both sonographers discussed their findings to reach a consensus. They confirmed the following US characteristics: the location, size, shape, boundary, pedicle, basal part, internal echo, whether containing cystic echo, marginal echo and rear echo changes as well as blood flow of the lesion.

The study was in accordance with the Code of Ethics of the World Medical Association. Approval was obtained from the Ethics Committee of Children's Hospital affiliated to Shandong University. Written informed consent was obtained from parents for the use of the data.

Statistical analysis

Continuous variables were expressed as a mean ± standard deviation or median according to the data distribution. Categorical variables were expressed as frequencies (percentages). Due to the small study population, no statistical tests were conducted.

Results

Clinical features

Among the 15 patients, 12 patients (80%) were males. Their ages ranged between 1 day to 12 years with a median age of two years. The patients presented with abdominal pain (n=10), vomiting (n=8), and hemafecia (n=7).

All 15 patients were treated with intestinal resection and anastomosis, and no complications were observed. Eleven (73.3%) lesions were located in ileum, approximately 30-100 cm away from the ileocecal valve with a median distance of 50 cm, two (13.3%) lesions in jejunum approximately 50 or 75 cm away from the ligament of Treitz, and two (13.3%) lesions in Meckel's diverticulum approximately 70 or 75 cm away from the ileocecal valve with the mass introverted at the top of Meckel's diverticulum. Surgeries demonstrated that the masses had a polypoid appearance with a diameter that ranged between 0.9-3.0 cm, with a median diameter of 1.4 cm (Table I).

Gross specimens showed that the mass was under the mucosa, and the boundary between the mass and intestinal wall was unclear. They often contain many small cysts, approximately 2-4 mm in diameter, with thick walls.

US features

Eleven patients were correctly interpretated by US as having adenomyoma, including nine ileal adenomyomas, one jejunal adenomyoma, and one Meckel's diverticulum. The diagnostic accuracy (the concordance rate between US diagnosis and pathological diagnosis) of small intestinal adenomyoma was 73.3%. Two (13.3%) patients were misdiagnosed, including one diagnosed as cystic masses in intussusception and one as intestinal duplication in intussusception. The other two (13.3%) patients were misdiagnosed, including one with coexisting small intestine atresia, and another with Merkel's diverticulum identified by US, but then failed to find adenomyoma.

In the 11 correctly diagnosed cases, most small intestinal adenomyomas had 1.0-3.0 cm with a median diameter of 1.3 cm, typically located in the submucosal region, with the basal part wide (100%) and long axis parallel to the attached intestinal canal without a pedicle, and the boundaries were clear (100%). The mass showed oval (72.7%) hypoechoic (90.9%) polycystic echo nodules, containing multiple small quasi-circular or irregular cysts of different sizes (81.8%) surrounded by solid hypoechoic mosaic areas, and the wall of the cystic areas was thick and uneven. The mass protruded into the intestinal cavity (81.8%), which may have resulted in local narrowing of the intestinal cavity with visible signs of intestinal content bypass. The color Doppler US showed in

Table I. Clinical findings and US diagnosis of 15 pediatric patients with intestinal adenomyomas

No.	Sex	Age	Symptoms	Ultrasound diagnosis	Location	Size (cm)	Distance from ileocecal / Treitz (mm)
1	F	2 d	H, vomiting	ileal mass with intussusception, intestinal obstruction	ileum	1.5/1.2/1.1	75
2	M	4 m	AP, H	adenomyoma of ileum with intussusception	ileum	1.4/1.0/0.9	75
3	M	12 y	AP	adenomyoma of ileum	ileum	1.9/1.6/1.1	100
4	M	4 y	AP	adenomyoma of ileum	ileum	1.1/1.0/0.8	50
5	F	1m	vomiting	adenomyoma of ileum with intussusception	ileum	1.2/1.0/1.0	50
6	M	5m	AP, vomiting	adenomyoma of ileum with intussusception	ileum	1.5/1.0/1.0	50
7	M	5 y	AP	adenomyoma of ileum with intussusception	ileum	1.7/1.2/1.0	100
8	M	1 d	H, vomiting	adenomyoma of ileum with intussusception	ileum	0.9/0.7/0.3	30
9	M	42 d	H, vomiting	adenomyoma of ileum with intussusception	ileum	1.2/1.0/0.9	75
10	M	2 y	H, AP, vomiting	adenomyoma of ileum with intussusception	ileum	1.6/1.2/0.8	75
11	M	7 y	AP	adenomyoma of jejunum	jejunum	2.0/1.2/1.1	50
12	F	7y	H, AP	Meckel's diverticulum with intussusception, intestinal obstruction	Meckel's diverticulum	1.1/1.0/1.0	75
13	M	8y	AP, vomiting	adenomyoma in Meckel's diverticulum with intussusception	Meckel's diverticulum	1.0/0.8/0.6	70
14	M	2d	vomiting	small intestine atresia	jejunum	1.1/1.0/0.5	75
15	M	7 y	H, AP	intestinal duplication with intussusception	ileum	3.0/1.2/1.0	50

F, female; M, male; H, hematochezia; AP, abdominal pain

the solid hypoechoic areas of the mass abundant (63.6%) or sparse(36.4%) blood flow signals (fig 1-4) (Table II).

Secondary intussusception was diagnosed in 11 (73.3%) patients using US as typical ultrasonic findings of "pseudo-kidney" and "target signs" of ileocolic intussusception, or multi-cystic mass within the distal end of the intussusception. Two patients with small intestinal adenomyoma in a Meckel's diverticulum caused secondary intussusception. Ultrasound images showed a gourd-like cystic structure with thick walls.

Discussion

In this study, we described the clinical features and US findings of small intestinal adenomyomas. Careful US examination showed a high diagnostic accuracy. This indicates that US is a possible and effective method for the examination of small intestinal adenomyomas. These findings are important because small intestinal adenomyomas could cause recurrent intussusception, and early detection and treatment can reduce the pain and radiation injury caused by recurrent intussusception and air enema. To the best of our knowledge, this is the first study to evaluate US diagnosis of small intestinal adenomyoma.

Adenomyomas occur at any age, including newborns and the elderly [9]. The most common locations were the stomach, duodenum, and biliary duct. However, they are

Table II. US features of 11 correctly diagnosed cases with intestinal adenomyomas.

Factors	US features	Frequency (n=11)
Location	Ileum	9(81.8%)
	Jejunum	1(9.1%)
	Meckel's diverticulum	1(9.1%)
Shape	Oval	8(72.7%)
	Round	3(27.3%)
Boundary	Clear	10(90.9%)
	Unclear	1(9.1%)
Pedicle	No	11(100%)
Basal part	Wide	11(100%)
Internal echo	Hypoechoic	10(90.9%)
	Isoecho	1(9.1%)
Marginal echo	Hypoechoic	7(63.6%)
	Isoechoic	2(18.2%)
	Hyperechoic	2(18.2%)
Rear echo changes	Hypoechoic	7(63.6%)
	Isoechoic	3(27.3%)
	Hyperechoic	1(9.1%)
Containing cysts	Several	9(81.8%)
	One	2(18.2%)
Protruding into the intestinal cavity	Yes	9(81.8%)
	No	2(18.2%)
Blood flow	Abundant	7(63.6%)
	Sparse	4(36.4%)

rarely found in the small intestine [5]. Additionally, adenomyomas of the small intestine are more common in the ileum than in the jejunum [32] and more common in boys than in girls. In our study, the location and sex ratio of intestinal adenomyomas were generally consistent with the data from 27 cases reported in the literature. The main manifestations of small intestine adenomyoma were vomiting, crying, abdominal pain, or hematochezia in younger children, and abdominal pain or hematochezia in older children. The small intestinal adenomyoma

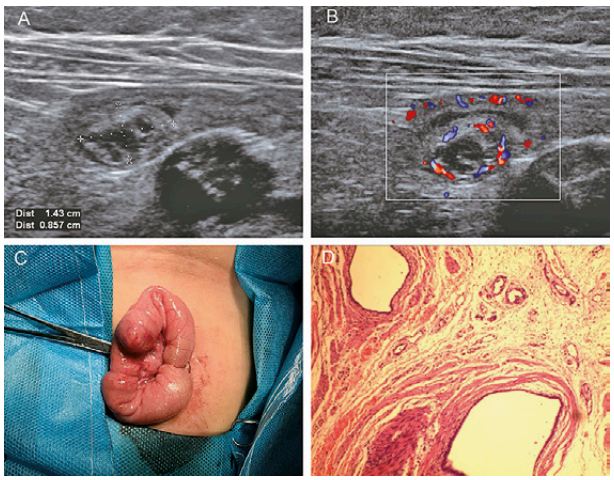


Fig 1. Ileal adenomyoma. A, Ultrasound imaging revealed a typical small intestinal polycystic mass in the right lower abdomen. B, CDFI showing vascularity in the wall of the cystic areas. C, Intraoperative photograph of the mass in the intestinal wall. D, Histological image with dilated glandular structures covered by monolayer cylindrical epithelium and surrounded by bundles of smooth myofibrils (hematoxylin and eosin stain, ×100).

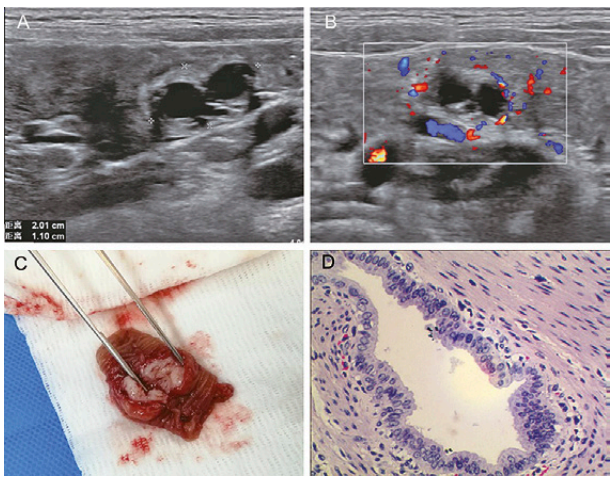


Fig 2. Jejunal adenomyoma. A, Ultrasound imaging of long-axis section of ileal adenomyoma in the left upper abdominal quadrant with the mass containing a polycystic structure. B, CDFI showing multipoint blood flow signals in the cystic wall. C, A gross section of the lesion is gray-white. D, Histological image showing bundled smooth muscle and dilated catheter (hematoxylin-eosin stain, ×400).

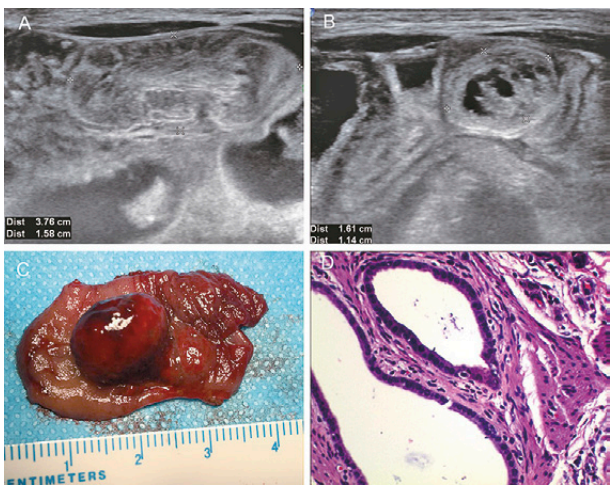


Fig 3. Small intestinal adenomyoma with intussusception. A, Ultrasound imaging of the upper abdomen revealed the typical ultrasonic finding of "pseudo-kidney" of the long axis section of intussusception. B, Transverse sonogram shows the mass (calipers) containing several irregular cystic areas. C, A dark red polypoid mass was seen on the section of the specimen. D, Histological image with the dilated glandular structures covered by monolayer cylindrical epithelium and surrounded by bundles of smooth myofibrils (hematoxylin and eosin stain, ×200).

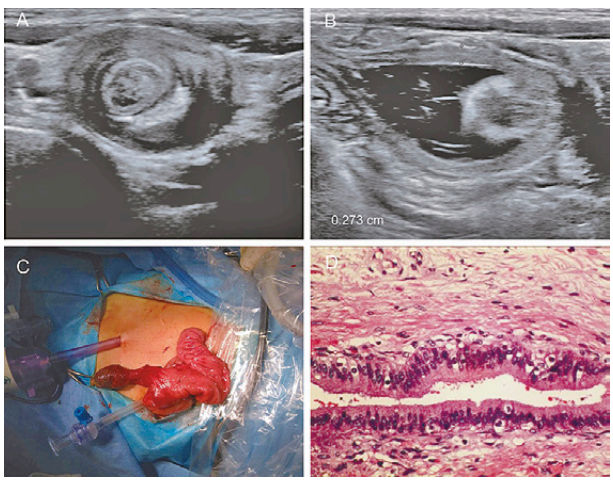


Fig 4. Small intestinal adenomyoma in a Meckel's diverticulum. A, Ultrasound imaging of the right lower abdominal quadrant revealed the typical ultrasonic "target sign" of ileocolic intussusceptions. B, Ultrasound imaging showing a structural of gourd-like cyst in sleeve. The wall of the cyst became obviously thicker, and solid hypoechoic nodules with enhanced marginal echo in the apical cavity of cyst. C, Intraoperative findings of the Meckel's diverticulum and its apical adenomyoma. D, Histological image with the dilated glandular structures covered by monolayer cylindrical epithelium and surrounded by bundles of smooth myofibrils (hematoxylin-eosin stain, ×400).

could cause recurrent intussusception, intussusception being the most common complication [1,3,15]. Adenomyoma-induced secondary intussusception and intestinal obstruction cause digestive system symptoms even intestinal necrosis.

We began to recognize this rare disease in a two-day-old newborn who underwent US examination because of crying and bloody stools. US revealed upper abdominal intussusception with intestinal obstruction. At the distal end of the intussusception, there was a clear oval, multilocular, hypoechoic nodule mass in the intestinal cavity without an obvious pedicle, which was different from the common Meckel diverticulum and intestinal duplication. We were very confused about this and followed up on the surgery and pathology examination. Pathological examination confirmed an ileal adenomyoma. Subsequently, we analyzed the US images of the children and found that the mass had oval hypoechoic nodules that tended to occur in the intestinal mucosa. The long axis of the tumor was parallel to the attached intestinal canal with clear boundaries, multilocular cysts, mosaic hypoechoic areas, and septations. Histologically, they comprise a mixture of dilated ductal structures lined by foregut-type cuboidal or columnar epithelium surrounded by irregular smooth muscle bundles [12]. Their epithelium has been occasionally reported to contain goblet and Paneth cells [10]. These ducts may secrete mucus components; therefore, some cystic elements are found in gross specimen [27].

According to our knowledge, no US description of intestinal adenomyoma, except for one case report of our group, was published [28]. Some case reports that had a definite diagnosis of intussusception were examined by US, but no adenomyoma mass was detected [4,25,26,29]. In our study, two patients were misdiagnosed, possibly because the child had a small intestine atresia or Meckel's diverticulum, with extensive dilation and obvious air accumulation in the proximal intestine, which made the identification of the adenomyoma difficult. There may be three reasons for misdiagnoses. First, doctors may lack knowledge on small intestinal adenomyoma. Second, US imaging could have been not clear due to obscuration by gas or stools. Third, most adenomyomas are small and inappropriate probe selection may lead to a lack of visualization. Therefore, negative US findings do not completely exclude the presence of intestinal adenomyomas. US examiners should improve the recognition of small intestinal adenomyoma, understand its US imaging features, choose the appropriate probe, follow the operational specification in the examination, and increase the pressure gradually on the gray scale to reduce the interference of gas or stool, to improve the detection rate of the lesions.

Although US can diagnose small-intestinal adenomyoma according to its typical ultrasonic features, attention should be paid to other nodular lesions in children. In our series of cases, one patient was misdiagnosed as intestinal duplication in intussusception. Intestinal duplication is usually located on the dorsal or mesenteric side of the maternal digestive tract segment, usually having a single lumen with no separation. US images show a spherical or tubular cystic structure with a "double-ring" or "triple-ring" sign on the wall, and the length varies from a few centimeters to the entire intestine [33]. Meckel's diverticulum is a remnant of the vitelline duct located at the contralateral margin of the mesentery [34,35]. The US features of Meckel's diverticulum are variable, with various shapes. It can be a typical diverticulum with a small amount of effusion or an atypical empty sac. It can also be expressed as a globular cyst or inflammatory mass. Juvenile polyps are the most common pediatric polyps and are usually solitary in the colon [36,37]. US images showed that juvenile polyps present as solid, hypoechoic nodules, with a variable number of small cystic that protrude into the bowel lumen and usually with a pedicle. Color Doppler US showed central blood flow in the pedicles and rich blood flow signals radiating from the center to the edge of the nodules. Polyps of Peutz-Jeghers syndrome, commonly found in the small intestine are also described as hamartomas [38]. US images showed that the polyp was a lobulated nodule with an alternative distribution of hyperechoic and hypoechoic regions.

Surgery is required for the definitive diagnosis of small intestinal adenomyoma and adenomyoma-induced secondary intussusception, including traditional methods and minimally invasive surgery with laparoscopy, which can achieve radical resection, while air compression enema and US-guided water enema are often the best choices for primary intussusception. The most important limitation in our study lies in its retrospective character from a single center. Secondly, because of the low incidence of this pathology, the number of patients included is relatively small. A multicenter study with appropriate sample size is needed to support our findings. Thirdly, a comparison between experienced and less experienced sonographers was not analyzed, and the intraobserver agreement was not assessed. Also, further studies need to be undertaken to demonstrate the feasibility and advantages.

Conclusions

Routine abdominal US is necessary when a child has abdominal pain, vomiting or/and hemafecia. If US finds an oval hypoechoic polycystic echo nodule located in the

submucosal region and protruded into the intestinal cavity, with the basal part wide, clear boundaries and without a pedicle, the small intestinal adenomyoma should be considered. US is an effective method for evaluating small intestinal adenomyomas and secondary intussusception in children.

Conflict of interest: none

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