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Case Report

Fatty-falciform ligament appendage torsion (F-FLAT): Diagnosis and management in a pediatric patient

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ABSTRACT

Fatty-falciform ligament appendage torsion (F-FLAT) is a rare condition manifested by torsion of the extraperiotoneal fat within the falciform ligament. It is similar to intraperitoneal focal fatty infarctions, including omental infarction and epiploic appendagitis. We report herein the first case of F-FLAT in a pediatric patient that failed conservative management. Ultrasound and CT scan facilitated prompt diagnosis of this rare finding and expedited a quick trial of conservative management. Despite conservative treatment, symptoms persisted for 4 weeks, but resolved after laparoscopic single-site surgical resection. In this report, we discuss the clinical features, key radiographic findings, and treatment options for this unique condition. This is only the third reported pediatric case of F-FLAT in the literature, and the first pediatric case to require surgical resection.

Clinical case

A 13-year-old girl presented to the emergency department with 3 days of epigastric abdominal pain. The pain localized to the mid-epigastrium and worsened with deep inspiration. The pain improved with abdominal flexion. On examination, her vital signs were within normal limits. Focal tenderness was elicited upon palpation of the epigastrium. Laboratory evaluation revealed a mild leukocytosis (14.2×10^9/L) (normal range $3.9\text{-}10.6 \times 10^9$/L). Basic metabolic and liver function tests were normal. Due to clinical concern for acute cholecystitis, a right upper quadrant ultrasound was performed.

In the region of the falciform ligament, between the left and right subphrenic spaces, ultrasound (US) demonstrated

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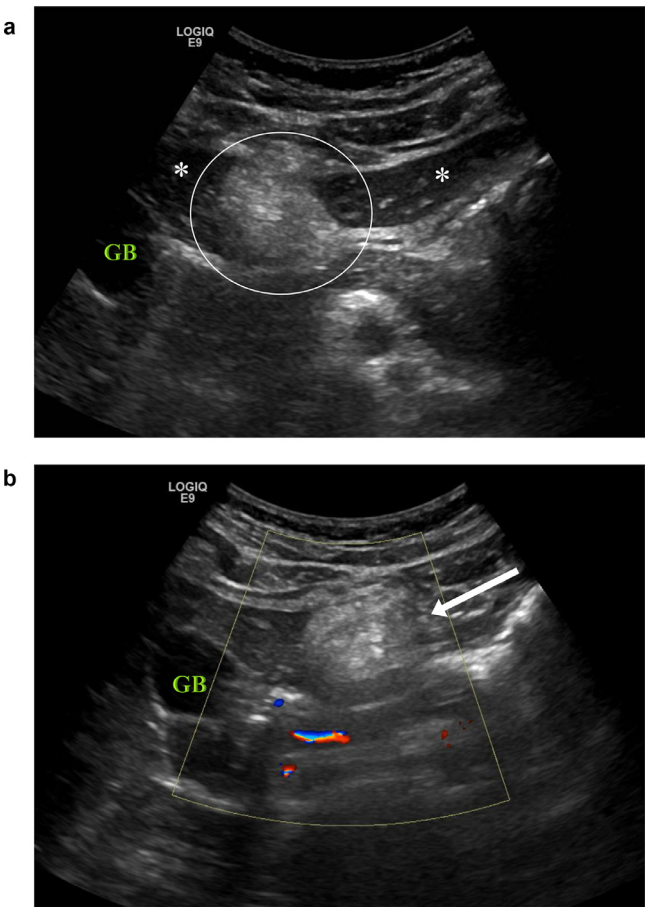

Fig 1 – (a) Transverse US demonstrates an ill-defined echogenic fatty mass (circle), in region of the falciform ligament diving the medial and lateral segments of the left lobe of the liver (asterisks). GB = gallbladder. (b) Transverse US mid abdomen with color Doppler. Image demonstrates a hyperechoic mass (straight arrow) with no internal flow. GB = gallbladder.

an irregularly marginated echogenic mass, void of spectral Doppler flow (Figs. 1a, b), measuring 5 cm in maximum AP dimension. Diffuse hypoechogenicity of the liver was also noted, suggesting an inflammatory process. A confirmatory CT scan of the abdomen/pelvis with IV and oral contrast revealed stranding of the peritoneal fat within the falciform ligament. The inferior extent of the fatty mass demonstrated a 'hyperattenuating rim' sign [1], an early sign of vascular occlusion. The vessels extending into the falciform ligament did not demonstrate contrast enhancement which substantiated concern for torsion of the fatty appendage of the falciform ligament (Fig. 2. a, b, c).

Surgical consultation was obtained, and conservative management was recommended via a trial of nonsteroidal anti-inflammatory medication' for the presumed diagnosis of F-FLAT. Despite 4 weeks of observation, the patient's pain persisted, and prevented her from returning to desired sporting activities. Ultrasound was repeated prior to surgery and demonstrated improved margination (Fig 3 a, b). Due

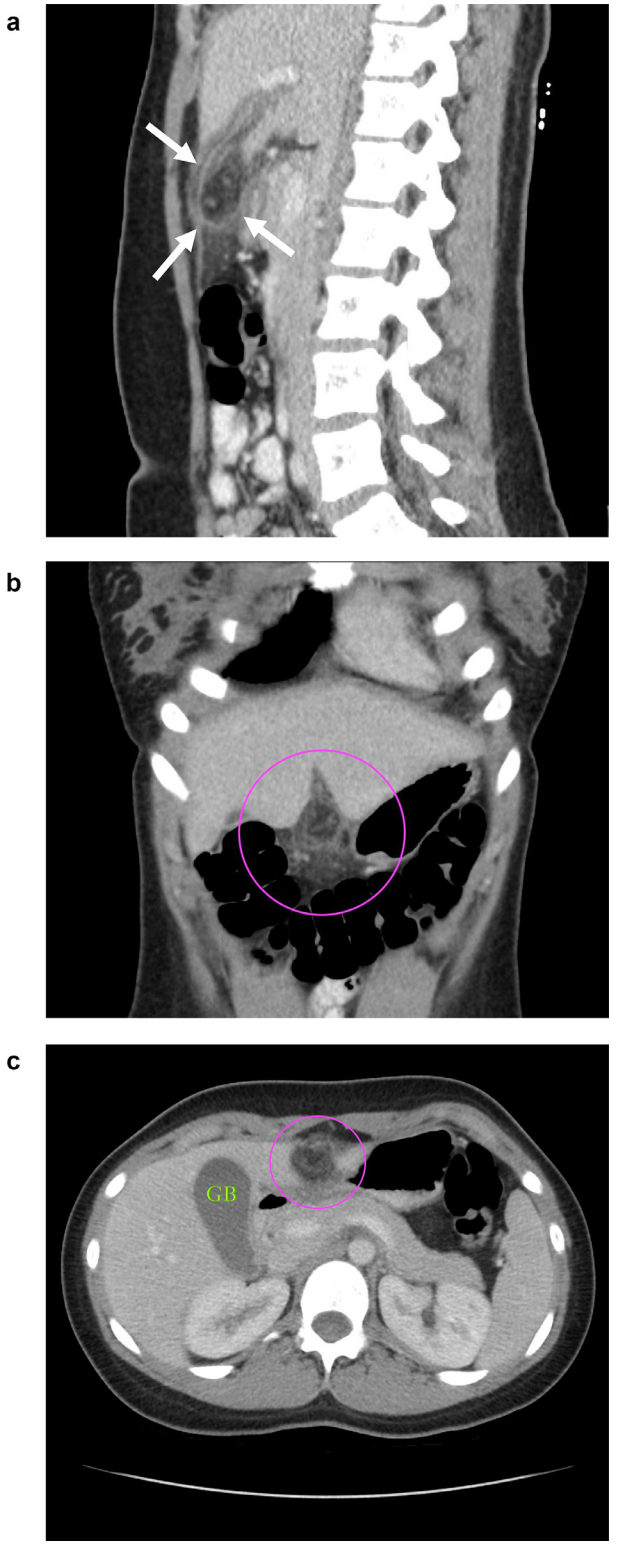

Fig. 2 – (a) Sagittal CT. Hyperattenuating rim sign and fatty mass with stranding extended into the liver along the course of the falciform ligament (arrows). (b) Coronal CT. (c) Axial CT. CT images of the abdomen and pelvis with IV and oral contrast. Coronal CT (b), and axial CT (c), demonstrate on oval fatty mass with a thin hyperdense rim along the course of the falciform ligament (circle). GB = gallbladder.

Fig 3 – (a, b) Two-week follow-up US imaging demonstrates decreased size and improved margination of mass indicative of decreased inflammation.

to persistent pain, the patient and her mother requested surgical excision.

A 2 cm incision was made through the umbilicus, through which a single-site laparoscopic port was introduced. A rubbery, fibrofatty soft tissue mass was identified in the falciform ligament, with extension into the round ligament of the liver (Fig. 4 a, b). The mass was resected with a laparoscopic bipolar electrosurgical instrument. Histology identified fat necrosis, vascular congestion, thrombosed vessels, and reactive mesothelial hyperplasia, suggestive of chronic inflammation. At a follow-up appointment, 2 weeks after the procedure, the patient's pain had resolved.

Discussion

F-FLAT is a very rare anomaly and may be confused clinically with other causes of abdominal pain. Focal epigastric pain in pediatric patients is far more likely to be related to inflammatory conditions such as acute gastritis, cholecystitis, or pancreatitis. A high index of suspicion is required to make this diagnosis and prevent children from undergoing unnecessary interventions, which can be often be avoided in lieu of conservative treatment. We will briefly review the key points to consider in the evaluation of this diagnostic oddity.

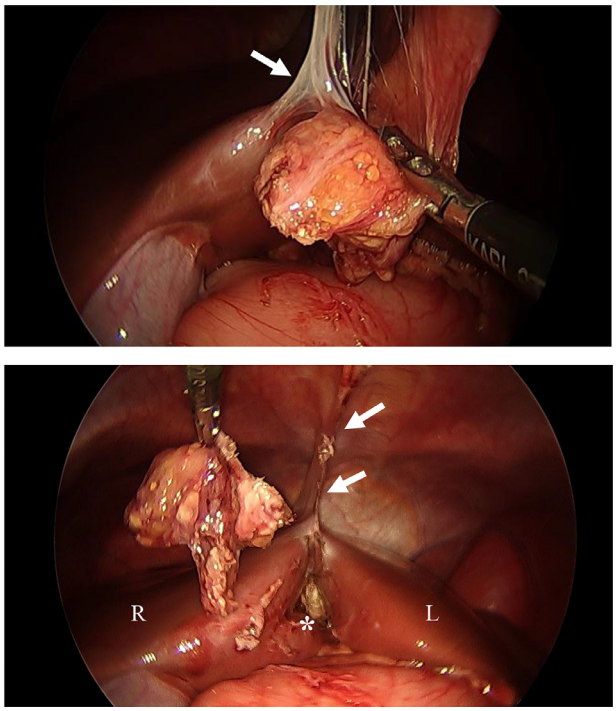

Fig. 4 – (a, b) Laparoscopic images of the fatty appendage of the falciform ligament show an enlarged fatty mass attached to the falciform ligament (a, arrow). Image b. F-FLAT resected from residual falciform ligament (arrows). R = right hepatic lobe. L = Lateral left hepatic lobe. * = Hepatic notch.

The falciform ligament is the remnant of the embryonic ventral mesentery. The anterior border connects to the peritoneum behind the right rectus abdominis muscle. It extends from the umbilicus to the superior diaphragm. The free edge contains the obliterated umbilical vein, or round ligament, which extends into the hepatic notch, and separates the medial and lateral segments of the left lobe. Conceptually, the course of the flaciform ligament when viewed sagittal has a semilunar footprint anterior to the liver. F-FLAT may occur anywhere along this track and should be scrutinized when presented with equivocal abdominal pain. The arterial blood supply primarily comes from the left inferior phrenic and middle hepatic arteries. Venous drainage of the falciform ligament usually flows into the left inferior phrenic vein [2]. The falciform ligament is unique in that it is a dual layer of visceral peritoneum with extraperitoneal fat separating the 2 layers. The extraperitoneal falciform fat can be variable in size. The fat can twist, thereby decreasing flow through its feeding vessels, and subsequently develop infarction, necrosis, and pain.

IFFI follow a clinical course very similar to F-FLAT, but originate from intraperitoneal, pedunculated structures such as the epiploic appendages of the colon, and the greater and lesser omentum. The narrow stalk and laxity of attachments of these structures may predispose to twisting. Infarction of the greater omentum has been reported in the pediatric population, more commonly so in obese children [3,4,5]. IFFI's may be found at the time of surgical exploration for abdominal pain of indeterminate nature and are commonly resected. However, if focal fatty infarctions are diagnosed radiographically, most will resolve with conservative treatment. Nonsteroidal anti-inflammatory drugs are the most commonly prescribed analgesics to control inflammation and pain, which usually resides within a week or 2 of diagnosis.

Diagnosing F-FLAT in a pediatric patient is very difficult for several reasons. Firstly, the diagnosis is extremely rare in children. To date, the radiographic diagnosis of F-FLAT has only been reported twice in the pediatric population. Maccallum et al. reported a 10-year-old boy who presented with 5 days of right-sided abdominal pain, vomiting, diarrhea, and anorexia. Ultrasound reported no abnormalities, but the diagnosis of torsion of a lipomatous appendage of the falciform ligament was made on CT scan and treated conservatively with oral analgesia [6]. Nam et al. reported a 13-year-old boy with 3 days of right upper quadrant pain. The diagnosis of torsion of the lipomatous appendage of the falciform ligament was made with both ultrasound and CT scan findings. The patient was treated conservatively, and experienced complete resolution of symptoms. Follow-up ultrasound imaging demonstrated near complete resolution of the previously seen torsed mass [7].

Currently, no reported cases of F-FLAT have been imaged with magnetic resonance imaging (MRI); all previous diagnoses have been made with ultrasound and CT. Ultrasound may be the initial diagnostic test of choice in pediatric patients with epigastric pain, given the lack of radiation of exposure. Sonographic findings suggestive of F-FLAT include a hyperechoic, noncompressible, slightly heterogeneous mass in the area of the falciform ligament. On real-time sonography, the fatty appendage may not be affected by respiration, which is suggestive of an extraperitoneal structure [8,9]. The typical CT finding of F-FLAT is an area of increased fat density, within or adjacent to the falciform ligament. The fat density extends into the epigastric region and is associated with surrounding inflammatory changes [8,9].

As many cases of F-FLAT are known to resolve without surgery, a period of conservative management with rest and nonsteroidal anti-inflammatory medication' is considered first-line treatment. Follow-up imaging with ultrasound, CT, or abdominal MRI may be considered if symptoms do not improve. Surgical resection of F-FLAT is generally reserved for cases in which pain persists and/or lifestyle is limited. The first identified case of F-FLAT requiring surgery was in an adult published by Webber et al. in 1977 wherein the torsion was an incidental finding during laparotomy of the patient with acute abdominal pain [10]. There is no consensus regarding the duration of nonoperative treatment prior to surgical intervention.

Conclusion

This report illustrates the pathophysiology, recommended diagnostic evaluation, and treatment of F-FLAT in the pediatric patient. The extreme rarity of F-FLAT mandates a radiographic evaluation, as the diagnosis has never been reported on clinical evaluation alone. Awareness of F-FLAT as a possible cause

of abdominal pain in the pediatric patient is an essential precursor to making the radiographic diagnosis. When identified, surgical intervention can often be avoided. This is the first reported case of F-FLAT in a female pediatric patient that did not respond to conservative treatment, and ultimately was cured with minimally invasive surgical resection.

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