Case Report

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A massive primary hydatid splenic cyst was successfully managed through open total splenectomy: a case report and review article

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Background: Cystic echinococcosis (CE) is a parasitic infection that is caused by the tapeworm Echinococcus granulosus. CE is very common, especially in the rural areas of developing countries. The most commonly affected organs by hydatid cysts are the liver and the lungs. However, the primary splenic hydatid cyst (PSHC) is a very rare manifestation of CE with an incidence of 0.5–8%.
Case presentation: A 17-year-old female patient presented with abdominal pain which gradually increased over months, along with anorexia and vomiting. Computerized tomography showed a massive splenic cystic mass. An open total splenectomy was performed. By follow-up, the platelet count and a postoperative chest X-ray were normal. The patient was prescribed Albendazole and analgesics. The pathological study confirmed the diagnosis of PSHC.
Conclusion: Despite the occurrence of PSHC being very rare, it is very important to take it into consideration, especially in endemic areas.

Keywords: case report, cystic echinococcosis, hydatid cyst, hydatidosis, large cyst

Background

Cystic echinococcosis (CE) is a parasitic infection affecting humans caused by the tapeworm Echinococcus species[1,2]. Mortal infection is incidental to intermediate hosts (in addition to lambs and scapegoats), which occur when humans ingest the parasite eggs present in the faeces of infected animals[1–3]. The original infection is asymptomatic, and the latent period can persist longer in most cases. It is generally discovered accidentally as a mass in the abdominal area in the left hypochondrium or lower common in the epigastrium or presented with pain, constipation, or dyspepsia[4]. CE affects the liver in 70% of cases, followed by the lungs in 25%, and the spleen in 0.5–8% of cases[1]. The opinion is generally grounded on clinical symptoms, radiological imaging, abdominal ultrasonography (USG), and computed tomography (CT), with or without serologic tests. The final opinion is verified by percutaneous aspiration and exploratory laparotomy in addition to histopathological examination[1]. The definitive treatment is splenectomy, either surgical or laparoscopic. In some cases, spleen-preserving surgery could be done[3]. This case has been reported in line with the SCARE criteria[5].

HIGHLIGHTS

- Despite hydatid disease being a common medical illness, primary splenic hydatid infection is a rare symptom, accounting for less than 3.5% of abdominal hydatid cases.
- The histological method is the most definitive method for diagnosis, along with other examinations such as abdominal ultrasound and computed tomography scanning.
- Surgery is the treatment of choice for splenic hydatid cysts.
- Splenic hydatid cysts should be suspected, especially in endemic rural areas.

Case presentation

A 17-year-old female patient residing in northeast Syria was admitted to the Department of General Surgery with a complaint of persistent upper left quadrant abdominal pain, vomiting, anorexia, and a gradual increase in abdominal size for 3 months. The patient had no relevant medical, family, or travel history. On clinical examination, she was vitally stable; by inspecting the abdomen, it seemed enlarged unsymmetrically; a mass occupying the upper left quadrant reaching the lower left quadrant was palpated, and it seemed to be dull on percussion. Her lab work

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was normal; both the haematological and metabolic panels were normal, and neither leukocytosis nor eosinophilia were noticed. The abdominal USG showed a fluid-filled, expanded stomach Figure 1A, while an enhanced CT scan showed a large, cystic spleen lesion (measuring 25 × 16 cm) with uniform fluid content, intact splenic texture, adjacent organ compression, and no free fluids in the abdomen Figure 1B. The differential diagnosis included a splenic hydatid cyst or a congenital cyst, but given the patient's geographical location, a preliminary diagnosis of a hydatid cyst affecting the spleen or the mesentery was made. An exploratory laparotomy was performed. Under general anaesthesia, an incision in the middle of the abdomen was obtained, which gave access to the abdomen.

On examination, the spleen occupied most of the abdominal cavity. It was occupied by a unilocular cyst measuring 18 × 15 cm. The gastrocolic, splenocolic, and gastrosplenic ligaments were transected. The splenic vessels in the splenic hilum were separated, transected, and secured to their central stumps with suture ligatures. The spleen was dislocated and removed.

There was no bleeding or other cysts. A drain was inserted, and the inspected field was lavaged. The abdominal wall was closed by sutures encompassing the fascia and peritoneum. The surgery lasted for an hour. The spleen was occupied by a unilocular cyst measuring 18 × 15 cm and weighing 5.9 kg, with a yellowish to white, trabecular, and ulcerated internal surface and a firm to hard wall surrounded by splenic tissue, which was ~2 cm thick (Fig. 2).

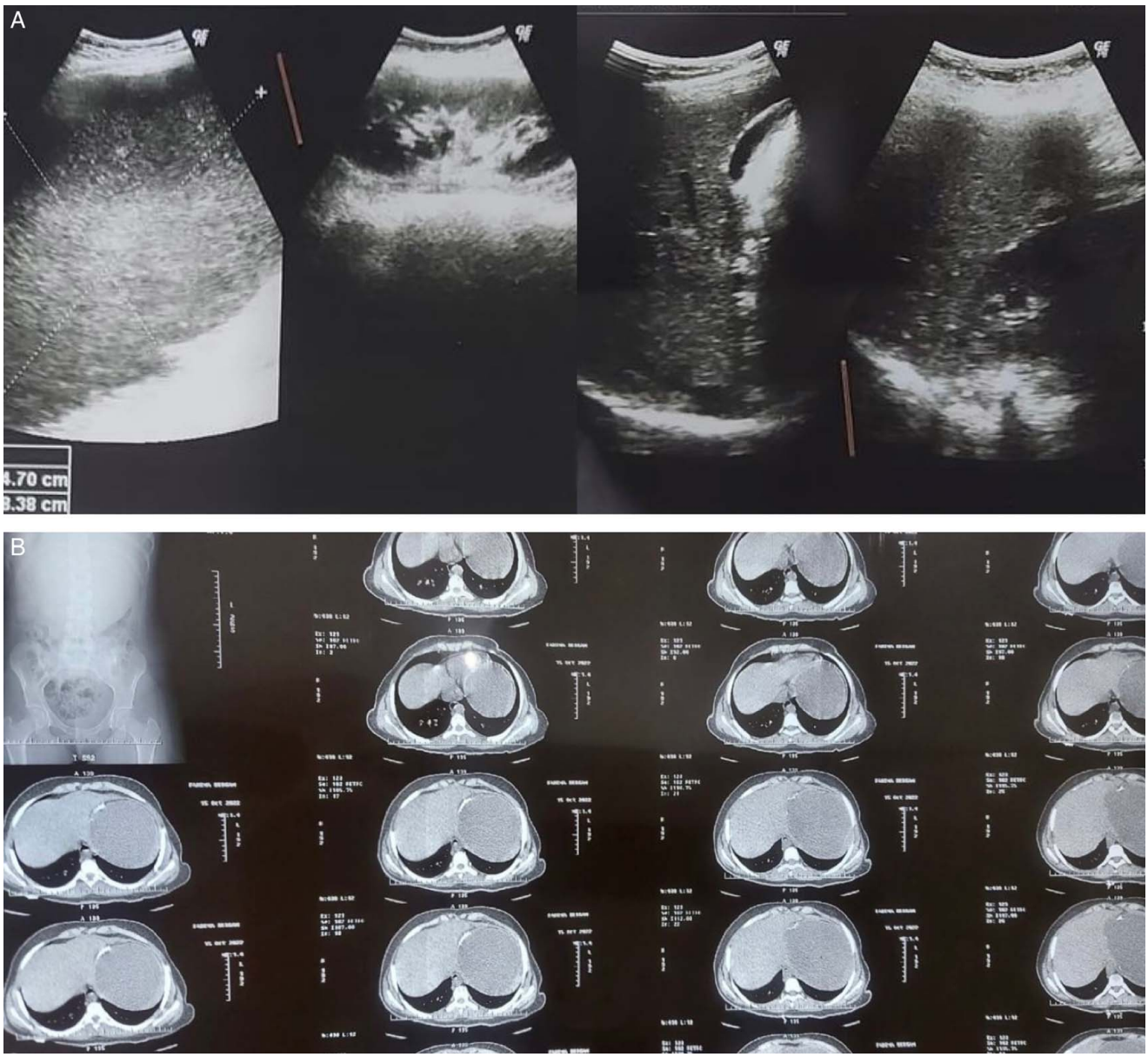

Figure 1. (A) Echography of the abdomen shows a giant lesion with pure fluids, the origin of the lesion is unrecognizable in this echography. (B) Axial images of an enhanced computed tomography scan of the abdomen show a large 25 × 16 cm, well-defined hypodense cystic mass of the spleen.

Figure 2. The spleen after splenectomy.

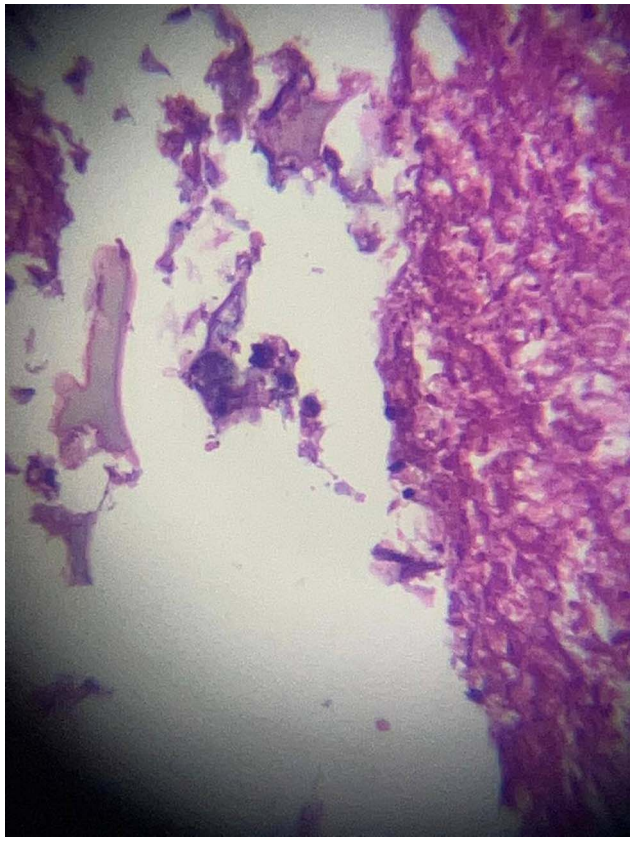

Figure 3. A section showing a vascular fibrocollagenous cyst wall with an inflammatory cell infiltrate and no epithelial layers. Multiple small fragmented acellular laminated membranes are noticed, matching a hydatid cyst membrane.

The cyst contained a serologic bloody fluid with no calcification or parasite tissue. The patient received pneumococcal, meningococcal, and Haemophilus influenzae vaccinations within 1-week post-operation because the decision of splenectomy was not definite before surgery. The hospitalization period was 3 days. Post-operation, the patient was in stable condition with no complications. The vital signs and laboratory findings were normal. The inserted drain output was steady in the 3 days. The patient was prescribed Albendazole for 3 months to avoid recurrence of splenic hydatid cyst (SHC), after the pathologhy report confirmed the diagnosis (Fig. 3), and analgesics to relieve pain.

Discussion

Berlot was the first to describe CE as a necropsy finding in 1790[4]. Primary hydatid cysts are a rare finding in the spleen, accounting for less than 3.5% of the entire abdominal hydatidosis. It is more common in rural areas. Especially in cattle-rearing areas of the Middle East, South America, South Europe, New Zealand, India, and Australia[4,6,7]. Our patient is a 17-year-old Syrian female from an endemic area, where northeast Syria is highly endemic for CE. The disease is common in both genders, most commonly in children, and the average age was 43.8 years (range 15–72 years)[7]. The prevalence of SHC is about 2.5–5%[3], with an incidence of 0.5–4%[4]. The occurrence ranges between 0.9 and 8[7], and between 0.5–8% of all hydatidosis[6]. There are many mechanisms to explain splenic involvement. The arterial route on which eggs can go beyond the liver and lung. The venous route is established by the retrograde spread of the worm via the portal vein to the splenic vein. And another route, like colonic trans-parietal and hepatic cyst rupture into the peritoneum cavity[3,4,7]. Our patient had a solitary splenic cyst. Consequently, the arterial or venous routes could apply to her case. By ultrasound and CT, cysts can be classified depending on Gharbi's classification (Table 1)[7]. Splenic hydatid cysts can be isolated or associated with other locations, most frequently the liver and lungs[1,6,7]. A few cases have demonstrated the association between splenic cysts and hypersplenism[4]. Our patient's cyst was solitary, without the involvement of other sites. The diagnosis of splenic hydatid disease (SHD) is recognized when a patient is assessed for other diseases[1,6,8–12]. Asymptomatic SHD repre-

Table 1

The classification of hydatid cysts based on US appearance

	Hydatid cyst classification according to Gharbi
Type I	Univesicular cyst
Type II	Detached membrane of the univesicular cyst (water-lily sign)
Type III	Multivesicular hydatid cyst
Type IV	Pseudotumoral hydatid cyst
Type V	Cyst with complete wall calcification

US, ultrasound.

sents ~30% of patients[1,6,8,11], but the diagnosis is based on clinical examination, imaging techniques, and serological tests[1,3,13,14]. SHD usually grows very slowly, often at a rate of 0.3–1 cm annually[6,15]. When the size of the cyst is large, patients can present with left upper quadrant pain and/or a palpable mass[1,6,9,12,13,16,17] and may present with nonspecific symptoms, including dyspnoea due to pushing up of the left diaphragm, dyspepsia, and constipation due to compression on the colon[8,11,17]. In our case, the patient was suffering from upper abdominal distension with nausea and vomiting without a history of any other disease. USG, CT, MRI, or serological tests can be used to evaluate hydatid cysts. The first-line method for detecting abdominal hydatidosis is USG. CT is superior to USG for identifying the cyst's number, size, and anatomic location; for identifying extrahepatic cysts; for spotting complications like infections, and for monitoring the lesions throughout treatment and detecting recurrences. MRI can identify rare cysts missed by USG and CT by providing cystic pathological features. However, because of its expensive price, it is not usually used. Serology makes a primary diagnosis and tracks treatment effectiveness[1,3,6,17]. The differential diagnosis is composed of other cystic formations of the spleen, including pseudocysts from post-splenic trauma, splenic abscesses, congenital cysts, metastatic disease, and cystic hemangiomas[3,8–11,13]. A variety of differential diagnoses and the rarity of SHD pose a diagnostic challenge. Radiological and serological tests (Indirect hemagglutination, enzyme-linked immunosorbent assay, or western blot) solve diagnostic dilemmas most of the time and reach the final diagnosis of SHD in 90% of cases[11]. Fine needle aspiration can assist in achieving a more definitive diagnosis or clarifying uncertain diagnoses[8].

Finally, during or after surgery, histopathology can confirm SHD. In our case, the USG was unremarkable, while the CT revealed a large-sized splenic cyst measuring 25×16 cm, which increased suspicion of SHD due to its presentation in endemic areas, and histopathology confirmed the diagnosis. Untreated SHD complications include secondary infection, inflammation, compression of other innards, bowel fistulization, and intra-abdominal cystic rupture of the peritoneal cavity, causing life-threatening peritonitis, which may lead to systemic anaphylactic shock[3,8,9]. There are many surgical and non-surgical approaches for SHD treatments, like total or partial splenectomy, spleen-saving approaches, puncture, aspiration, injection, respiration (PAIR), and medical treatment[2,4,12,18]. Total splenectomy is the standard treatment because it has low mortality (the related mortality rate ranges between 1.9% in adults and 4% in children) and morbidity rates and decreases the rate of complications of cyst rupture[18]. A splenic cyst with adhesions or infiltrations to nearby organs, a large cyst (more than 75% of the spleen parenchyma), multiple, symptomatic cysts, or a hilar location should be treated by total splenectomy[4,8,9]. The most common complications are haemorrhage, gastric and pancreatic injuries, subphrenic abscess, thromboembolic, chest infection, and overwhelming post-splenectomy infection, especially in children under the age of 4[3,19]. Also, the laparoscopic approach has not been recommended due to the high risk of peritoneal spillage[20], but it is usually performed for patients with unique, small-sized, superficially located cysts[7], taking advantage of its minimally invasive nature. The morbidity rates for laparoscopic and open approaches were 14.2% and 33.3%, respectively[4]. As spleen-saving approaches, there are cysts enucleation, unroofing with omentoplasty, and partial splenectomy. Splenic preservation using these methods or PAIR treatment should be attempted if possible. Spleen-saving approaches are used for patients at a young age with small, solitary, and peripheral location cysts[4,18,21]. With spleen-saving approaches, some complications may occur. For example, there is a risk of poor vascular control when incising splenic tissue, residual cavity infection, haemorrhage, and subsequent intra-abdominal abscess. There was no significant difference between the splenectomy and spleen-preserving groups concerning median hospital stay, postoperative complications, or recurrence rate[9,22]. The PAIR is the only diagnostic and therapeutic method[23], and is advocated in cases with surgery contraindications or high anaesthesia risk, and contraindicated in uncooperative patients and mature calcified cysts and is less successful for multiseptated cysts that contain numerous daughter cysts and cysts that on sonography appear to be mostly solid[3]. Small and simple types of hydatid cysts may be treated with PAIR alone or in combination with Albendazole. With PAIR, there could be some complications, like haemorrhage, anaphylaxis, hypersensitivity reactions, pneumothorax, and pleural effusion as early complications and infection in the cavity, recurrence, and dissemination as late complications. Also, percutaneous treatment of splenic CE has a low rate of morbidity, short hospitalization, and uncertainty about the potential recurrence rate[22]. These patients should be closely followed up with serology and sonography. Signs of cure, like a decrease in the dimensions of the cysts, solidification of the contents, and irregularities in the walls of cysts, signs of healing should be seen[3]. Medications are recommended for 1-month pre-surgical and post-surgical procedures with Albendazole and Praziquantel[3]. It helps to sterilize the cyst, reduce the tension in the cyst wall, decrease the chance of anaphylaxis, and reduce the recurrence rate postoperatively[8]. Also, for small (< 5 cm) and asymptomatic cysts, patients who have complications and who refuse surgery or are surgical non-candidates may only be treated with Albendazole[8]. Approximately 30–50% of patients have shown improvement with medical treatment. However, medical treatment should never be relied upon as a substitute for surgery because 25% of patients did not show any improvement, and even the majority of patients who responded to treatment experienced a recurrence after 2 years of discontinuing the therapy[3]. Preoperative or early postoperative vaccination helps minimize post-splenectomy sepsis risk, especially in highly endemic and poor countries; Prolonged postoperative anticoagulant therapy, antibiotherapy, and a complete blood count every 2/week are convenient choices[3,8], (Table 2). In our case, the total splenectomy procedure was used due to the cyst's huge size. The patient was discharged 3 days postoperatively and was prescribed Albendazole and analgesic drugs, and no sign of recurrence was found.

Conclusion

The diagnosis of SHC is usually doable, based on serology and imaging, especially for those living in or travelling to echinococcosis-endemic regions. For large cysts, total splenectomy is suitable (> 10 cm) with a high risk of rupture or compression of other vital structures. It may have high morbidity and mortality, but it is recommended in poor and underdeveloped countries. A conservative procedure is safe, but it may also result in postoperative collection and a higher recurrence rate. To ensure complete healing, post-surgical pharmacological treatment is necessary.

Table 2

Characteristics, Diagnosis, and management of reported cases of Splenic cysts

Paper N	Age (year)/ sex of pt	Main complaint	Diagnostic tests	Findings	Tx and FU
1	44/F	Left hypochondrium pain	USG—CT—Pathology	USG: anechoic solitary, thin-wall cyst in splenic parenchyma. CT: well-defined cystic lesion in upper pole of spleen pathology: acellular fibrous wall, scolex of hydatid cyst	laparoscopic splenectomy (3 months)
4	44/M	Dull abdominal pain, bloating under L rib cage	USG—CT—Pathology—Blood tests	USG: anechoic solitary, thin-wall cyst in splenic parenchyma. CT: well-defined cystic lesion in upper pole of spleen pathology: acellular fibrous wall, scolex of hydatid cyst	laparoscopic splenectomy (Per 6 months/4y)
5	52/M	Dry cough, fever, night sweats	USG—CT—MRI—Pathology—Blood tests	Blood tests: slight eosinophilia MRI: enlarged spleen containing a well-defined capsulated cyst CXR: abnormal lucency in left upper quadrant of the abdomin	laparoscopic splenectomy (4 months)
7	C1: 30/M C2: 27/M	C1: loss of vision, headache, painless, progressive protrusion of the right eye. C2: fever, abdominal swelling in the left hypochondrium, dragging sensation	C1: Blood tests—CT—FNA—CXR—Abd USG C2: Blood tests—CXR—Abd USG	C1: eosinophilia—CT: intraorbital well-defined, hypoechoic, cystic lobulated mass, focal calcifications—FNA: clear fluid, scolices of echinococcus granulosus C2: USG: uniloculated cystic mass in upper pole of spleen with patchy calcifications in cyst wall.	C1: enucleation of eyeball C2: splenectomy
8	28/M	Mass in the left abdominal upper quadrant, Left hypochondriac dull aching pain	CT—Pathology	CT: large homogenous cystic lesion in spleen	Splenectomy
9	35/F	Left upper abdominal pain	USG—CT—explorative laparotomy	USG: large well-defined round cystic mass in epigastric and left hypochondrium CT: large cystic space occupying lesion arising from spleen explorative laparotomy: large splenic cyst	Splenectomy
10	28/F	Asymptomatic	CT—USG—Pathology	CT\USG: well-defined complex cystic lesion	Spleen-preserving surgery
11	75/F	Abdominal pain in left hypochondrium	CXR—CT—Blood tests	CXR: elevation of left diaphragm Blood tests—haemoglobin, mild leukopenia and eosinophilia CT: cystic lesion in upper pole of spleen	Laparoscopic splenectomy (3months)
12	49/F	Chronic pain in left upper quadrant	USG—CT—Pathology	USG: uncertain cystic mass, solid components CT: septated splenic cyst Pathology: parasitic cyst	Splenectomy (6 months)
13	16/F	Pain in the left hypochondrium	USG—CT—Pathology	USG: bulky hydatid cyst CT: large well-defined cystic lesion involving almost entire spleen Pathology: hydatid cyst	Splenectomy
14	41/F	Abdominal pain	USG	USG: large splenic cyst	Splenectomy
15	49/F	Abdominal pain and distension	Blood tests—CT—Pathology	Blood tests: eosinophils CT: splenomegaly, nonenhancing cystic lesion, Hepatomegaly, gross ascites, portal hypertension	Emergency laparotomy, splenectomy
16	29/M	Abdominal pain	CT—USG—Pathology	CT: relative splenomegaly, well-defined complex cystic lesion Pathology: active hydatid cyst	Splenectomy (3 months/1 year)
19	C1: 44/M C2: 71/F	C1: nonspecific abdominal pain. C2: satiety after meals, abdominal discomfort	C1: CT—USG C2: USG—CT—Serologic tests	C1: Abdominal USG: enlarged spleen. Abdominal CT: splenic calcified hydatid cyst C2: Abd USG: calcified splenic mass CT: circumferentially calcified spleen + Serologic tests: E. granulosus	C1: laparoscopic splenectomy (3months) C2: splenectomy
21	27/M	Asymptomatic	USG—CT—MRI	USG:splenomegaly CT—MRI: cystic mass in spleen	PAIR method

Abd, abdominal; CT, computed tomography; CXA, Clivoaxial angle; F, female; FNA, fine needle aspiration; FU: follow-up; M, male; PAIR, puncture aspiration injection respiration; Tx, Treatment USG, ultrasonography.

Ethical approval

Not applicable.

Consent

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

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Author contribution

M.M.: design of the study, data collection, data interpretation and analysis, drafting, critical revision, approval of the final manuscript. K.N.: design of the study, data collection, data interpretation, and analysis, critical revision, drafting, approval of the final manuscript. M.M.A.: data interpretation and analysis, critical revision, drafting, approval of the final manuscript. M.K.: drafting, critical revision, approval of the final manuscript. F.A.: drafting, critical revision, approval of the final manuscript. Y.A.: drafting, critical revision, approval of the final manuscript. H.S.: drafting, critical revision, approval of the final manuscript. Y.A.: The co-supervisor, drafting, critical revision, approval of the final manuscript. R.S.: The co-supervisor, drafting, critical revision, approval of the final manuscript. O.K.: The Supervisor, patient care, drafting, critical revision, approval of the final manuscript.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

Research registration unique identifying number (UIN)

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Data availability statement

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