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TORSION OF A WANDERING SPLEEN (a case report)

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Abstract---*Introduction. Splenic dystopia, or a wandering spleen, is a rare and unusual case which is represented by movability of the spleen from its normal left hypochodrical position to other pelvic cavities. It appears as a result of many factors such as congenital insufficiency of the ligamentous apparatus, atypical location and elongation of its vascular pedicle. But the laxity of the spleen's primary supporting ligaments is considered to be the most agreed-upon factor. This rare pathology affects mainly young adults, particularly women and children, which presents as an asymptomatic abdominal mass or abdominal discomfort due to the torsion of the pedicle. Clinical manifestations are presented as asymptomatic up to abdominal emergency. Treatment is mostly surgical. The aim of this work is to present and analyze the anamnesis and the treatment of the case. Presentation of case: A case report of splenic torsion from the surgical department of the 3-clinic of the Tashkent Medical Academy is described in the present work. This is a 20 year-old lady with complaints for pain in the left hypochondrium, nausea, dry mouth, forced position due to the pain and general weakness. The patient underwent the following studies: an irrigoscopy – for the Hirschsprung's disease, MSCT of the abdominal cavity, laparoscopic splenectomy. Surgery "Laparotomy. Splenectomy. Drainage of the abdominal cavity" was performed. Review of literature was made using the keyword combination: "splenic torsion". Discussion: A splenic torsion is either congenital or acquired. Laparoscopic approach is the preferred surgical intervention. Conclusion: The diagnosis of splenic torsion is very rare, extremely difficult to establish and is clinically nonspecific, especially in patients presenting with acute abdomen. An early diagnosis and surgical care are required for preserving the spleen. Additional imaging examinations (ultrasonic scan and CT) are predominant and can help establish a correct diagnosis, before development of life-threatening complications. Surgery is often necessary and Plenectomy is preferable.*

Key words---*splenic dystopia, wandering spleen, torsion, splenectomy.*

I. INTRODUCTION

The case of splenic dystopia (wandering spleen) is rather interesting and rare pathology. There is not enough information on the observation of this disease in Uzbekistan, and only about 500 cases reported worldwide, where the incidence rate of 0,2% [1]. Splenic dystopia is a disease caused by congenital insufficiency of the ligamentous apparatus

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and atypical location of this organ. The most severe complication of splenic dystopia is the torsion of the splenic vascular pedicle, which can lead to splenic infarction and rupture [2, 10, 12].

One of the first documented descriptions of splenic dystopia was made by Dr. Josef Dietl, a Polish clinician, who not only documented three cases between 1854 and 1863, but also described the laxity of splenic ligaments as an etiology [3].

Thus, this pathology leads to mobility and migration of the spleen to other plenic cavities that in its turn causes the torsion of the elongated splenic pedicle. Splenic dystopia is managed surgically with a laparoscopic splenectomy.

The spleen is fixed by ligaments, which are the folds of the peritoneum directed to the hilus lienis (Fig. 1).

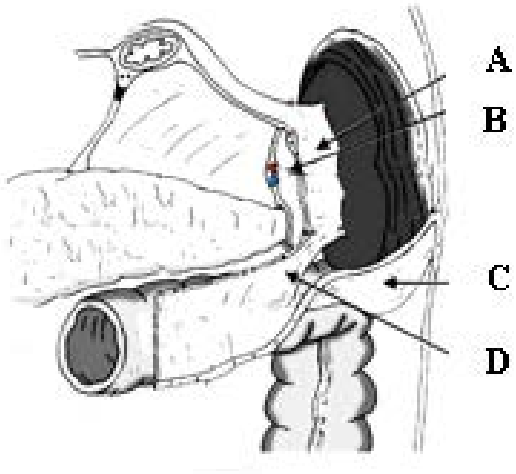

Fig. 1. Ligaments of spleen: A – lig. gastrolienalis; B – lig. lienorenalis; C – lig. phrenicoocolicum; D – lig. lienocolicum.

Knowing the anatomy of the spleen ligaments helps at the performance of surgical interventions on spleen, stomach, pancreas and colon.

Presentation of case

A lady A.M., 20 years old, was admitted to the surgical department of the 3-clinic of the Tashkent Medical Academy on June 14, 2019 with diagnosis: “Primary: Acute thrombosis of the splenic artery and vein. Complications: Pain syndrome. Concomitant: Chronic anemia of 1st degree ".

The patient is a native of Tashkent, temporarily unemployed. From the anamnesis, she considered herself sick for 2 months, when had felt a pain in the left hypochondrium, nausea, dry mouth, forced position due to the pain, and general weakness. She had an outpatient treatment with a temporary pain relief effect. For the last 10 days the abovementioned symptoms intensified, after which she was examined. A splenomegaly, a spleen hemangioma and ascites were revealed by the ultrasonic scan. Besides, the patient was performed an irrigoscopy – for the Hirschsprung's disease and MSCT of the abdominal cavity, where a dense formation under the left dome of the diaphragm, 11x9.5x4.5 cm in size, soldered to the small intestinal loops (lymphoma), splenomegaly and thrombosis of the splenic vein were defined. In this regard, on 12.06.2019 she passed examination at the Republican Specialized Scientific and Practical Center of Oncology and

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Radiology, where her oncological pathology was excluded. With the complaints mentioned above, finally she addressed to the 3-clinic of TMA and was hospitalized in the surgery department.

At admission to the clinic the general condition is moderate severe, skin and visible mucous membranes are pale, body temperature is 37.4 ° C. No eruption on skin and visible mucous membranes. Peripheral lymph nodes are swelled.

A weak breathing is heard from both sides in the lungs, without wheeze. Respiration rate is 18 per minute. At auscultation of the heart the tones are clear, clean and rhythmic, the heart rate is 86 per minute. Blood pressure is 120/80 mm Hg. Art. The tongue is dry, covered with a white fur. By the white line on the abdomen, a post-surgical scar 25x0.5cm of size is defined (the patient bore a surgery on January 15, 2019 "Sigmoid colon reversal. Mesosigmoplication by Hagen-Thorne. Transanal intubation. Sanitation and drainage of the abdominal cavity" with a diagnosis “Dolichosigma. Introversion of sigmoid intestine. Diffuse serous peritonitis"). The abdomen is of usual form, asymmetrical, because of the left half swelling; engaged in breathing act, soft, painful in the left hypochondrium. Peritoneal symptoms and muscle tension of the anterior abdominal paries are not marked. Liver and spleen are not enlarged. No peripheral oedema. Stool and diuresis are normal, regular.

Blood group and Rh factor: B (III) Rh + (positive).

General blood test by 06/14/2019: hemoglobin 94 g/l, erythrocytes 3.5 * 10^{12}/l, leukocytes 7.0 * 10^{9}/l.

Urinalysis by 06/14/2019: the reaction is acid, protein and glucose - negative, the epithelium - 0-1-2-3/1, leukocytes - 1-2-3/1, salt - ++.

Biochemical blood test by 06/14/2019: alanine aminotransferase 30,0 U/L, aspartate aminotransferase 26,0 U/L, bilirubin 8.3 mmol/l, urea 6.03 mmol/l, creatinine 74.0 mmol/l, total protein 66,0 g/l.

Coagulogram dated 06/14/2019: fibrinogen 3.60 g/l, prothrombin index 94%, international normalized ratio 1.17, hematocrit 36%, thrombotest 6, VSC 3’40”-4’00”.

Ultrasonic scan by 15.06.2019: signs of the spleen infarction, with possible abscess formation (the spleen 278x112 mm, of biconvex shape with rounded edges, the upper third of the spleen is a heterogeneous rounded formation 133x91 mm of size.

The patient passed the examination of a cardiologist, where no cardiological pathology was revealed, after which she was recommended a treatment of the primary disease.

The following treatment was performed at the hospital: an infusion therapy - Glucose sol. 5%-400 ml (in-v), Sodium chloride sol. 0.9%-400 ml (in-v), Adexin sol. 200 ml (in-v), Sepid sol. 250 (tr-f); for improvement of blood rheology – Sorbitol sol. 200 (in-v); for resolvent purpose – Diclofenaci sol. 3.0 (in-m); for analgetic purpose – Analgini sol. 50% -2.0 + Dimedroli sol. 1% -2.0 ml (in-m); for antibacterial purpose - Cephtriobak 1.5 + Novocaini sol. 0.5% -5.0 (in-m), Lefloksoli sol. 100 ml (in-v).

Considering the pain in the left hypochondrium and increasing it in the dynamics the patient was made a preoperative diagnosis on June 17, 2019: “Primary: An acute thrombosis of the splenic artery and vein. Complications: Splenomegaly. Spleen infarction. Pain syndrome. Accompanying: Chronic anemia of 1st degree”. The surgery “Laparotomy. Splenectomy. Drainage of the abdominal cavity” was performed. The spleen is absent in the left subphrenic cavity. The spleen was located in mesogastric over the descensive colon, 15x12x9 cm of size, of black color and dense consistency;

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torsion of pedicle is observed at hilus lienis (Fig. 2, 3). Splenectomy was performed. The sigmoid colon is dilated, swelled in shape (Fig. 4).

Postoperative diagnosis: «Primary: Splenic dystopia. Complication: Torsion of the spleen pedicle. Splenomegaly. Splenic necrosis. Pain syndrome. Accompanying: Dolichosigma. Adhesive disease of the abdominal cavity. Chronic anemia of 1st degree».

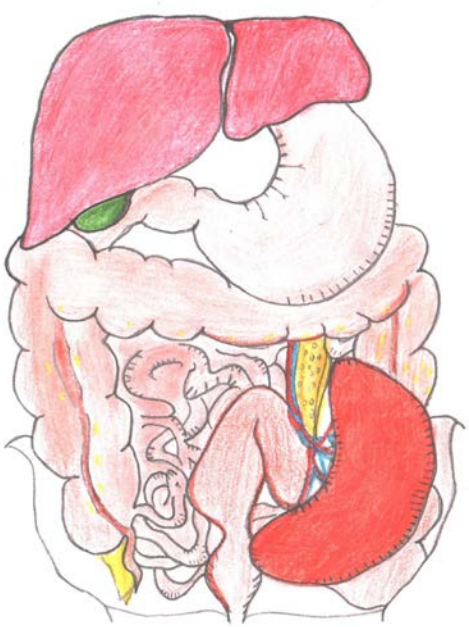

Fig. 2. Schematic image of the splenic dystopia with the torsion of pedicle.

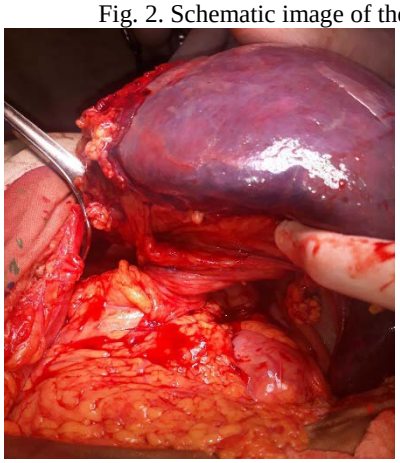

Fig. 3. Splenic dystopia with the torsion of pedicle;

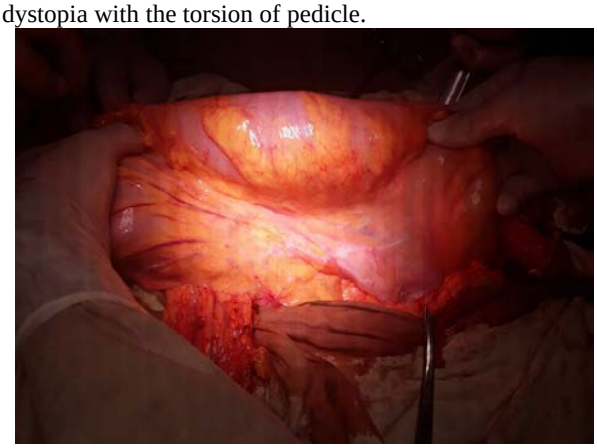

Fig. 4. Dolichosigma

The postoperative period proceeded without complications, a course of antibacterial, infusional and analgetic therapy was performed. Besides it, the patient underwent a blood and plasma transfusion on one dose. In the dynamics Hemoglobin is 90 g/l. On the control ultrasonic study - on the echograms of the abdominal cavity the presence of free limited humor was not found. In the projection of the spleen bed the presence of a residual cavity is absent. The postoperative wound was kept clean and healed by first intention. On the 4th day after the surgery period, the drainage tube was removed from the abdominal cavity. Histological examination by 06.17.2019: splenic necrosis.

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The patient underwent postoperative antibacterial, infusional and analgetic therapy. She was signed out from the department in a satisfactory condition.

II. Review of literature

As it was said above splenic dystopia is one of the rarest clinical finds and is observed in less than 0.5% of cases of the total number of splenectomy, in which an abnormal location of the spleen in the abdominal cavity or in the small pelvis is revealed.

Soleimani M. jointly with co-authors performed an advanced literature search on this issue and found out that from 1895 to 2005 there were described 238 cases of splenic dystopia, most of which are presented like descriptions of isolated clinical cases. In the available literature review from 1895 to 2016 there are 454 observations of patients with such pathology [4].

Forms of the splenic dystopia are various. The spleen may be located transversely under the left half of the diaphragm, including the cavity between the stomach arch and the diaphragm, as well as may be in the umbilical hernia, in the retroperitoneal space. When the internal organs are reversed, the spleen is on the right side, not the left. At the displacement of the spleen into the pelvis, it must be clearly distinguished from the ovarian tumor. In addition, the case of spleen prolapsus into the scrotum is described. After finding the splenic dystopia it is necessary to check whether the spleen is really missing in its proper place so that to exclude the possibility of two or multiple spleens. It is known that the surplus (or accessory) spleens are not rareness. But mostly the accessory spleen is small and located in the hilus lienis of the main spleen, along the splenic vessels in the gastrosplenic ligament. Although, there are cases when a quite large accessory spleens are localized in the caul, in the mesentery of the transverse colon, in the kidney capsule, in the tail of the pancreas, in the Douglas space, and etc. [5].

There are cases when 20-45 accessory spleens were found in the abdominal cavity. Finally, the spleen tissue may be found in the parenchyma of another organ. At weakness of the ligament apparatus, the spleen becomes movable ("wandering" spleen). The reasons of this can be both congenital and acquired factors. Congenital causes are hypoplasia of the ligament apparatus of the spleen, particularly the splenic-colon and gastro-splenic ligaments. Acquired causes are injuries and pregnancies accompanied by high levels of estrogen. The majority of the described cases of the “wandering” spleen are characterized, as a rule, by asymptomatic or scarce symptoms, including subacute abdominal pains and typical gastrointestinal symptoms - complaints to nausea, vomiting and constipation. However, in the case of the splenic torsion, the symptoms of the acute abdomen appear. The torsion of the vascular leg of the spleen leads to its infarction and necrosis. In clinical practice, idiopathic splenic torsion is very rare, so the precise diagnosis is established, as a rule, only in the process of a surgery. In some cases, the splenic dystopia can cause a compression of the adjacent organs of the digestive tract and lead to the development of ileus. [3].

In 2015 V.N. Kolocey and V.P. Strapko described a clinical case with acute intestinal obstruction. The patient was appointed a set of conservative actions aimed at treatment of acute intestinal obstruction [5]. The diagnosis of acute intestinal obstruction did not raised doubts, and in connection with the results obtained during the physical examination, an ileocecal invagination was suggested as a possible cause of obstruction [5]. The performed therapy had no success, so

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therefore the patient was offered a surgical treatment. Splenic dystopia and soldered loops of the small intestine on the inner surface of the spleen were found out intraoperatively.

As a rule, uncomplicated forms of splenoptosis have no clinical manifestations and turn to be a diagnostic finding at ultrasonic study, magnetic resonance and multispiral computer tomography, radionuclide scintigraphy. According to indicators angiography or diagnostic laparoscopy can be performed [2].

The method of the surgical treatment choice for splenic dystopia is splenectomy. In some cases, when finding the "wandering" spleen, including the torsion of the vascular pedicle, detortion and splenopexy are carried out. Such kind of surgery is recommended for young patients, taking into account the risk of post-splenectomy sepsis [1, 3].

Discussion. Very likely that the spleen is the least understood and the most discredited entity among all vital organs in the human body. Limited literature is found regarding splenic dystopia, mostly case reports. Generally splenic dystopia complicated with the torsion is met at children under 10 years old and young adults 20-40 years old, females at most. The documentation of the first case on this pathology goes to the Polish clinician, Dr Jozef Dietl. He not only revealed the life-threatening complications of this condition, but also investigated the reasons and factors of the spleen dystopia.[2]

Splenic dystopia being a rarely diagnosed case is either congenital or acquired pathology. [7,8] Congenital splenic dystopia is secondary to the failure of fusion between the dorsal mesogastrium and posterior abdominal wall during foetal development [7]. The acquired one is generally met in reproductive age of young women, particularly pregnant ones, which can lead to ligamentous lengthening due to laxity of the abdominal wall and hormonal changes in the body, as well as due to traumas.

The spleen has six peritoneal attachments (primary suspensory ligaments) that are directly associated with it (gastrosplenic, splenorenal, splenophrenic, splenocolic, pancreaticosplenic and presplenic folds) and two ligaments (pancreaticocolic and phrenicocolic) in indirect association. Failure of fusion of the dorsal mesogastrium to the posterior abdominal wall during embryogenesis leads to failure or defective attachment of these ligaments, leading to splenic dystopia. The gastrosplenic, splenorenal and phrenicocolic ligaments have been primarily implicated. (Fig.2)[11]

Fig.2

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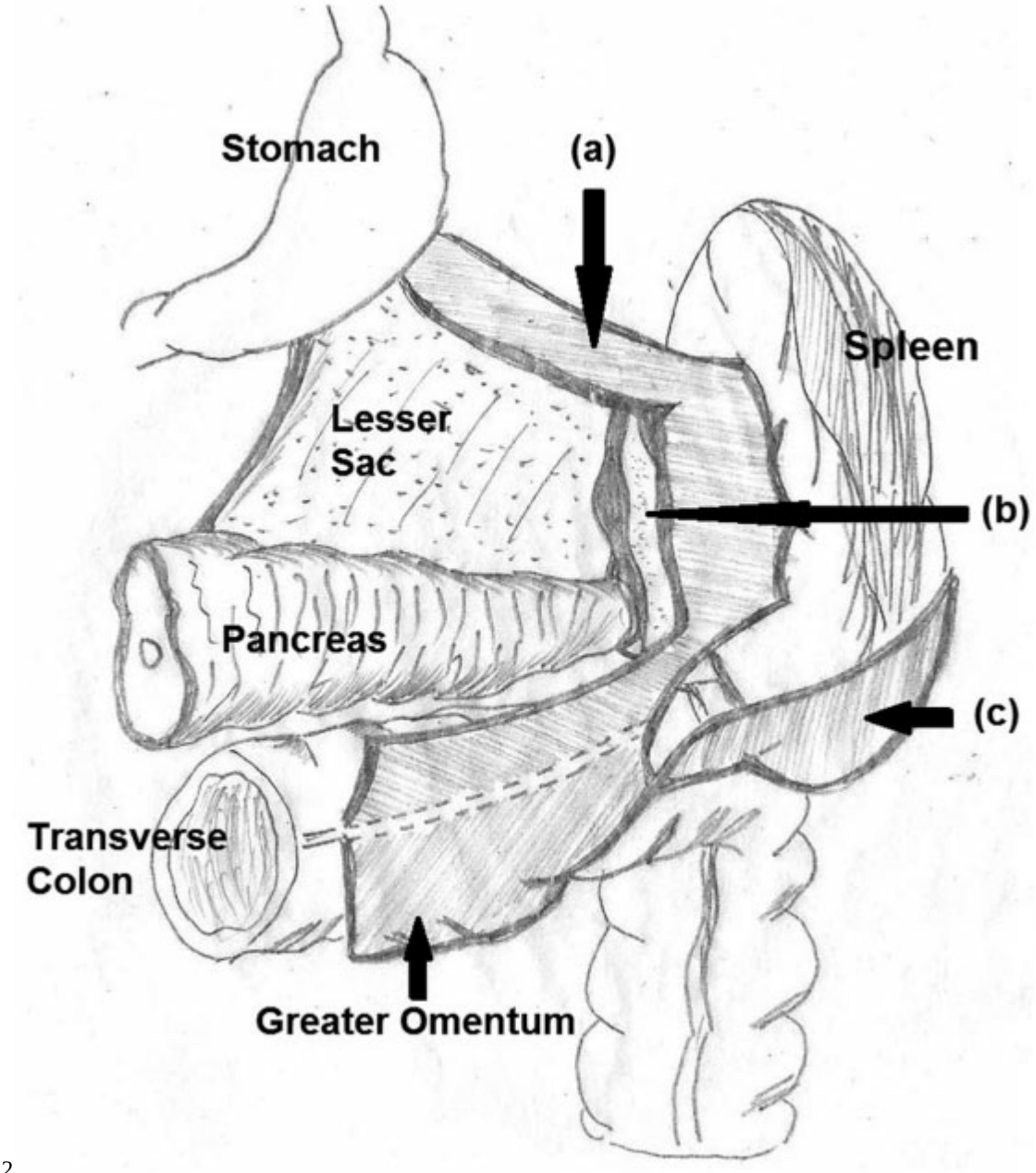

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There are some conditions associated with splenic dystopia: enlargement or absence of kidney, infectious mononucleosis, malaria, Hodgkin's disease, Gaucher's disease and previous pregnancy.

Symptoms of the disease may remain absent for long periods of time. As a result, the abnormal position of the spleen predisposes the splenic vascular pedicle to become tortuous, elongated and prone to intermittent torsion, in turn making the spleen vulnerable to a partial or complete infarction.

The surgical intervention is inevitable. A patient with splenic infarction due to torsion of the splenic pedicle, as in the presented case above, is offered splenectomy. Splenoplexy, either open or laparoscopic, is offered to most of the other

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patients in whom splenic pedicle detorsion and splenic fixation to either the diaphragm or abdominal wall are done [9]. Non-operative treatment of splenic dystopia is not advised as there is a 65% chance of torsion with ischemic splenic infarction without fixation of the spleen [10]. A clinician should have a reasonable suspicion for splenic dystopia, particularly in women of child-bearing age and children who present with acute abdominal pain and a mobile abdominal mass. Modern imaging techniques (CT, ultrasonic scan, Doppler US and etc.) are usually diagnostic and can identify the splenic pedicle torsion with a high degree of accuracy. Surgical operation in the form of splenectomy is largely governed by the findings of pedicle torsion and the associated risk for acute splenic infarction [9].

Complications related to the torsion of spleen are quite common and may cause progressive splenic infarction and necrosis. If the torsion is progressive, results in infraction of arterial supply, acute ischemia, strangulation, necrosis and splenic rupture. However, splenic infarction is, undoubtedly, the most common complication.

III. Conclusion

Thus, the splenic torsion at a wandering spleen is considered to be a rare pathology in the abdominal surgery and requires an urgent surgical intervention. Applying of instrumental methods of researches allows to effectively resolve the issues of preoperative diagnosis and observation of the patient after splenectomy.

Torsion of wandering spleen is extremely difficult to establish without primary tests and ultrasonic scans. Though it has quite importance for differential diagnosis in patients who present with acute abdomen. Its diagnosis should be made promptly before life-threatening complications.

The best method of confirming the diagnosis is CT scan and Doppler US imaging.

An early diagnosis and surgical care are required for preserving the spleen, especially in children. Laparoscopy usually confirms the diagnosis.

De-torsion and splenopexy is the method of choice, whenever possible because, in addition to conserve spleen, prevent any future complications. When there is an infarction spleen, splenectomy is necessary [10].

So, optimal treatment requires a high level of reasonable suspicion, early diagnosis and prompt surgical intervention, where preservation of the spleen is the goal.

Conflicts of interest. The authors declare that they do not have any personal conflicts of interest.

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Ethical approval. Being this present study a paper review, it is exempt from prior evaluation by the ethics committee.

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).

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