CASE REPORT

Single-port Splenectomy after Splenic Cyst Aspiration for Huge Splenic Cyst with High CA 19-9 Levels: The State of the Art

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ABSTRACT

Aim: Description of a patient with an elevation of CA 19-9 levels caused by a giant benign splenic cyst, which was completely laparoscopically resected through a single port, despite the large size of the lesion. An increase in CA 19-9 in this type of cystic tumors does not indicate malignancy.

Background: The cystic tumors of the spleen are rare diseases that may present a CA 19-9 increase, without indicating malignancy. There is a tendency to perform surgical intervention as less invasive and aggressive as possible. The current trend in the management of splenic cysts with a CA 19-9 increase and benign radiological characteristics, is a minimally invasive surgery, whenever possible.

Case description: We present a case of a young patient with a giant splenic cyst of 20.5 × 14.5 × 23 cm and elevated CA 19-9 levels. Spleen was completely laparoscopically resected through a single port after the cyst puncture and aspiration, using the advantages of laparoscopic surgery and single port. Had a favorably postoperative course with resolution of the symptoms up to the current date.

Conclusion: There is an actual tendency to perform minimally invasive surgical interventions. In pathologies such as the clinical case provided, we should try to make a surgical intervention as less invasive as possible, despite the large size of the lesion. In our case, we successfully performed the resection of a giant splenic cyst, using the advantages of laparoscopic surgery and the single port, which helped us in the extraction of the piece.

Clinical significance: When facing this pathology, we must bear in mind that benign epithelial and mesothelial cysts can produce an increase of CA 19-9 blood levels, without indicating malignancy. So, we should try to make a surgical intervention as less invasive as possible.

Keywords: CA 19-9, Laparoscopy, Mesothelial cyst, Single-port, Splenic cyst.

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BACKGROUND

Splenic cystic tumors are rare diseases, with an estimated incidence of 0.07%. Benign epidermoid and mesothelial cysts may present a CA 19-9 increase, without indicating malignancy, which is why they usually cause the clinician to be confused. We present a case of a young patient with a giant splenic cyst and elevated CA 19-9, which was completely laparoscopically resected through a single port after the cyst puncture and aspiration.

CASE DESCRIPTION

A healthy 19-year-old woman with no personal history of interest presented pain in the left hypochondrium of 2 months of evolution accompanied by dyspepsia, anorexia, and weight loss. On physical examination, a palpable mobile mass was observed. Abdominal CT (Fig. 1) showed a splenic cyst of 20.5 × 14.5 × 23 cm with a marked mass effect towards the stomach without infiltration of adjacent organs. Blood analyses were normal, without portal hypertension or hypersplenism data, but CA 19-9 blood level of 2,496 U/mL was highlighted.

Because of the patient's symptoms, a laparoscopic splenectomy was performed through a single transumbilical port, extending the incision to the supraumbilical midline about 3 cm. As the first step, the umbilical trocar ring was introduced, and a controlled aspiration of the cyst content was made prior to splenectomy. After aspiration and cyst collapse, the hermetic seal of the single trocar

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was locked, and pneumoperitoneum performed. The pericystic adhesions were released with the help of the ultracision (Fig. 2B) and subsequent section of short vessels. The vessels of the splenic hilum were sectioned with endoGIA (Covidien, USA) 60 mm vascular load (Fig. 2A), and the piece was extracted through the single port.

Definitive pathological anatomy of the surgical specimen was a benign epidermoid cyst (Fig. 3). The cytology of the cyst content was negative for malignancy.

After the surgical intervention, the patient progressed favorably, with resolution of the symptoms up to the current date. Analysis performed after the intervention showed normalization of CA 19-9 levels.

DISCUSSION

Splenic cysts are rare diseases, with an estimated incidence of 0.07%. The incidence is higher in young women between 18 years and 46 years.

They are classified as parasitic and nonparasitic. The nonparasitic are subdivided into true or primary cysts (25%) and pseudocysts or secondary cysts (75%) depending on the presence of a coating of epithelial cells inside the cyst.¹

Fig. 1: Abdominal CT shows the splenic cyst of 20.5 × 14.5 × 23 cm with a marked mass effect towards the abdominal organs

They are usually asymptomatic, but when they are large, as in our patient, can produce symptoms such as abdominal pain, postprandial discomfort due to compression of the stomach, sensation of a palpable mass, hemorrhage, infection or spontaneous rupture.²

The diagnosis is usually done with imaging tests: Abdominal ultrasound shows the cystic lesion that may have septa inside; CT scan can show calcifications in the cystic wall; the NMR reveals a hyperintensity signal in the T1 and T2 sequences.³ In addition, 11% of cases are diagnosed when a complication occurs, such as bleeding, rupture, and infection.

CA 19-9 is a glycoprotein produced in the epithelial cells of the ducts of the salivary glands, pancreatic duct, bile, and metaplastic mesothelial cells. It can be elevated in gastrointestinal, pancreatic, and biliary carcinomas, so it is used as a tumor marker, although we can find false positives in benign diseases such as cholangitis, pancreatitis, liver cirrhosis, cystic fibrosis, and idiopathic pulmonary fibrosis.⁴

An increase in CA 19-9 level does not indicate malignancy in this type of cystic tumors, although the surgeon must be alert to dismiss the possibility of a cystic pancreatic tumor. There are similar cases reported in the literature with a CA 19-9 blood level normalized after splenectomy and the anatomopathological result of benignity.⁵ The epithelial cysts can elevate this tumor marker without a correlation between the size of the cyst and

Figs 2A to C: (A) The release of pericystic adhesions. The arrow points to the silk thread of the aspiration point; (B) The section of the splenic hilum with endoGIA (Covidien, USA) 60 mm vascular load; (C) Single postoperative scar. H, splenic hilum; S, spleen; white line, marks the outline of the splenic cyst

Figs 3A and B: (A) Macroscopic pathological anatomy; (B) Microscopic image of the splenic cyst (hematoxylin-eosin): the wall of the cyst is lined by mature nonkeratinized squamous epithelium without atypia

CA 19-9 blood levels.⁶ For this reason, the current trend for these tumors with a CA 19-9 increase is the least invasive surgery instead of laparotomic access given the benignity of the similar cases reported with a CA 19-9 elevation.⁵

Differential diagnosis should be made with congenital cysts, infection by parasites, cystic neoplasms of the tail of the pancreas, and previous splenic trauma.⁷ There are described some cases of primary splenic cystadenocarcinomas⁸ and splenic lymphomas⁹ that course like a splenic cyst. Although they usually look like solid lesions, hemorrhagic phenomena and necrotic degenerations can cause them to acquire a cystic appearance.¹⁰

Surgical treatment is indicated when they are symptomatic, have a size greater than 5 cm or complications appear.¹¹ The gold standard is total splenectomy¹² although, partial splenectomy, marsupialization or fenestration of the cyst, can also be performed.¹³ Partial splenectomy could be performed for maintaining immunity against encapsulated bacteria although, the incidence after splenectomies has decreased due to vaccination against these pathogens;¹⁴ however, this procedure presents some risks such as intraoperative and postoperative hemorrhage and cystic recurrence.¹⁴ For asymptomatic splenic cyst smaller than 5 cm, a close follow-up is recommended, since cases of spontaneous cystic regressions have been described.¹⁵

There are cases of splenic cysts resected by open laparotomy or by laparoscopic access if they are not large.¹⁶ Laparoscopic splenectomy has proven to be a safe procedure, with advantages over open approaches, such as a reduction in hospital stay, less postoperative pain, and a faster postoperative recovery.¹⁷

Therefore, the current trend in the management of splenic cysts with a CA 19-9 increase and benign radiological characteristics is minimally invasive surgery whenever possible. In this case, we adopted a combined approach with the aspiration of the cyst and subsequent splenectomy by a single port, which becomes a valid approach for most of the cysts with these characteristics. In the literature review, we have not found splenic cyst cases handled with the help of the single port.

CONCLUSION

There is a tendency to perform surgical interventions less invasive due to the lower surgical aggressiveness and a decrease in the associated morbidity. In pathologies such as the clinical case provided, we should try to make a surgical intervention as less invasive as possible, despite the large size of the lesion. In our case, we successfully performed the surgical intervention of a giant splenic cyst, using the advantages of laparoscopic surgery and a single port, which helped us in the extraction of the piece.

CLINICAL SIGNIFICANCE

When facing this pathology, we must bear in mind that benign epithelial and mesothelial cysts can produce an increase of CA 19-9 blood levels, without indicating malignancy. So, we should try to make a surgical intervention as less invasive as possible.

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