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CASE REPORT

A Rare Complication of Atraumatic Splenic Rupture in Infectious Mononucleosis: A Case Report

Paschalis Gavriilidis[1,*], Pantelis Xanthakos[1]

[1]Department of Surgery, General Hospital of Syros, "Vardakeio and Proïo", Hermoupolis, Syros, Greece
*Correspondence: pgavrielidis@yahoo.com (Paschalis Gavriilidis)

Abstract

Aims/Background Atraumatic splenic rupture is a very rare and potentially life-threatening event usually associated with underlying pathological conditions. Splenic rupture in infectious mononucleosis occurs only in 0.1%–0.5% of patients. The aim of the present study was to publish a case report of the atraumatic splenic rupture and to present a mini-review of the international literature.

Case Presentation A 30-year-old man of mixed races (Caucasian and African) diagnosed with acute cholecystitis was referred by a rural doctor to a secondary district hospital. His past medical and family history was unremarkable. He did not report any allergies. On arrival, his vital signs were stable. However, laboratory examinations demonstrated: white blood cells 26 × 10^3/μL (4–11), neutrophils 38.8% (35%–72%), lymphocytes 58% (20%–45%), red blood cells 3.59 M/μL (0–0.6), haemoglobin 10.9 g/dL (13.5–17.5), haematocrit 33.4% (40%–54%), platelets 11.5 fL (6.5–11), prothrombin time 13.2 s (9.4–12.5), glucose 70 mg/dL (74–107), sodium 135 mmol/L (137–146), calcium 7.6 mg/dL (8.8–106), serum glutamic-oxaloacetic transaminase 426 U/L (10–45), serum glutamate pyruvate transaminase 530 U/L (7–45), gamma glutamyl transferase 151 U/L (7–49), total albumins 5.3 g/dL (6.6–8.3), C-reactive protein 235 mg/L (<5), and Epstein-Barr virus 15.50 S/CO (0–1.0). In addition, computed tomography determined hepatosplenomegaly, haemoperitoneum, and spleen rupture. Physical examination revealed abdominal rigidity, left shoulder tip pain, shortened capillary refill time, and neck lymphadenopathy.

Results The patient underwent expeditious total splenectomy, postoperative period was uneventful and he was discharged on the sixth postoperative day. He was scheduled to undergo the post-splenectomy vaccinations and regular follow-ups with his general practitioner abroad.

Conclusion In patients without a history of trauma, spontaneous splenic rupture should be considered in the differential diagnosis if patients have complaints of abdominal and left shoulder tip pain, and laboratory results indicate low haemoglobin and haematocrit levels.

Key words: spleen; spontaneous; atraumatic splenic rupture; infectious mononucleosis; case report

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Introduction

The most common cause of damage to the spleen, an organ responsible for hematopoiesis in the abdomen, is forceful abdominal trauma. 93% of patients with atraumatic splenic rupture are attributed to pathological causes, whereas idiopathic causes account for 7% of cases (Khan and Jindal, 2010). Atraumatic rupture in infectious mononucleosis (IM) is a very rare complication, occurring only in 0.1%–0.5% of patients (Farley et al, 1992). Usually, patients with (IM) present with a

clinical picture of the triad pharyngitis, lymphadenopathy, and fever. However, all patients (100%) demonstrate splenomegaly on ultrasound examination. Approximately 50% of these patients exhibit clinical splenomegaly (Dommerby et al, 1986). Moreover, 90% of them demonstrate hepatitis with mildly elevated serum transaminase levels (Schuler and Filtzer, 1995).

The cardinal symptoms of splenic rupture include left upper quadrant pain or tenderness, left shoulder tip pain (Kehr sign), abdominal distension, syncope, and hypotension. Abdominal ultrasonography at the bedside can detect a blood volume of 100 mL and has a sensitivity of 90% for detecting traumatic splenic rupture (Ma et al, 1995). Computed tomography (CT) is suitable for haemodynamically stable patients and provides assessment of other abdominal organs; moreover, intraparenchymal haematomas are easier to identify, and consequently, delayed splenic rupture can be managed accordingly (Clancy et al, 2012; Gamblin et al, 2005; Hyun et al, 1972).

Both traumatic and atraumatic causes are classified. The most common cause of traumatic ruptures is motor vehicle injury with a prevalence range from 50%–75% (Clancy et al, 2012). Atraumatic splenic ruptures are very uncommon; however, when they occur, they have a mortality rate of 12%–20% (Kocael et al, 2014; Renzulli et al, 2009).

Atraumatic splenic rupture (ASR) is defined by the Orloff and Peskin (1958). These criteria are as follows: (1) detailed history not revealing any history of trauma, (2) no evidence of disease in any organs apart from the spleen, (3) no evidence of perisplenic adhesions or scarring consistent with previous trauma history, and (4) a macroscopic and histological examination that shows the spleen to be normal (Orloff and Peskin, 1958). In 1991, Crate and Payne (1991) added a fifth criterion; they stated that no increase in antibodies, suggestive of viral infection in the acute or convalescent phase, should be confirmed, and they considered viraemia as a cause of rupture. The pathological causes of ASR can be divided into seven categories: infectious, haematological, neoplastic, inflammatory, iatrogenic, primary splenic, and idiopathic (Table 1) (Franklin and Casós, 2006; Hildebrand et al, 2014; Howdieshell et al, 2006; Kaldas and Gavriilidis, 2019). Infectious diseases and neoplasia account for more than half of all cases (Hildebrand et al, 2014).

Depending on the degree of injury, management can be divided into conservative, splenic artery embolisation, or splenectomy (Franklin and Casós, 2006; Howdieshell et al, 2006). It is crucially important that patients who undergo splenectomy are vaccinated with haemophilus influenza type B, pneumococcal, and meningococcal group C conjugate vaccines (Gavriilidis et al, 2020; Howdieshell et al, 2006; Skattum et al, 2012). However, there is no solid evidence supporting the need for vaccination of conservatively managed patients (Davies et al, 2002).

Oral phenoxymethylpenicillin and erythromycin are recommended lifelong prophylactic antibiotics (Jibawi and Cade, 2010). In particular, local evidence-based protocols recommend a minimum of 2 years prophylactic antibiotics for adults in general and lifelong for patients with sickle cell disease and lymphoproliferative disorders (Jibawi and Cade, 2010).

Table 1. Causes of atraumatic splenic rupture.

Neoplastic	Haematological	Infectious	Inflammatory	Primary splenic disorder	Iatrogenic
Lymphomas	Haemolytic anaemia	Epstein-barr virus	Rheumatoid arthritis	Splenic infarctions or venous thrombosis	Thrombolytic therapy
Leukaemia	Idiopathic thrombolytic purpura	Cytomegalovirus	Lupus erythematosus	Splenic angiomatosis	Anticoagulant therapy
Myelodysplastic disorders	Haemophilia	Human immunodeficiency virus	Primary amyloidosis	Splenic cyst	Dialysis
Multiple myeloma	Factor XIII deficiency	Hepatitis A/B/C	Acute/chronic pancreatitis	Splenic peliosis	Lithotripsy
Polycythaemia vera	Protein C deficiency	Varicella	Polyarteritis nodosa	Portal hypertension	Granulocyte-colony stimulating factors
		Rubella		Congenital malposition (i.e., short splenic pedicle)	Colonoscopy
		Bacterial Infective endocarditis Legionella Bartonella Malaria Syphilis Toxoplasma			

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The purpose of the present study was to present the complication of atraumatic splenic rupture in infectious mononucleosis. Google Scholar, Embase, Pubmed, Medline and Cochrane Library systematically searched for pertinent literature. Consequently, based on the above results mini review was written.

Case Report

A 30-year-old man of mixed races (African and Caucasian) was referred by a rural doctor to a remote secondary hospital for acute cholecystitis. The patient's medical history was unremarkable. His past medical and family history was unremarkable. He did not report any allergies. His vital signs were stable upon arrival at the hospital (blood pressure, 118/88 mmHg; cardiac beads 84; oxygen saturation 98%; and temperature 36.3 °C). Physical examination revealed generalised abdominal rigidity, left shoulder tip pain, reduced capillary refill time, hepatomegaly, splenomegaly, and neck lymphadenopathy. The patient reported having sore throat pain.

Laboratory results were: white blood cells 26 $\times$ 10^3/μL (4–11), neutrophils 38.8% (35%–72%), lymphocytes 58% (20%–45%), red blood cells 3.59 M/μL (0–0.6), haemoglobin 10.9 g/dL (13.5–17.5), haematocrit 33.4% (40–54), platelets 11.5 fL (6.5–11), prothrombin time 13.2 s (9.4–12.5), glucose 70 mg/dL (74–107), sodium: 135 mmol/L (137–146), calcium 7.6 mg/dL (8.8–106), serum glutamic-oxaloacetic transaminase 426 U/L (10–45), serum glutamate pyruvate transaminase, 530 U/L (7–45), gamma glutamyl transferase 151 U/L (7–49), total albumins 5.3 g/dL (6.6–8.3), C-reactive protein 235 mg/L (<5), and Epstein-Barr virus 15.50 (0–1). CT revealed splenic rupture, large hemoperitoneum, and hepatosplenomegaly (Fig. 1). The repeated complete blood count examination demonstrated a further decrease in haemoglobin from 10.9 to 8.5 g/dL and Hematocrit (HCT) from 33.4% to 25.8%.

The patient underwent expeditious laparotomy revealing 2000 mL of hemoperitoneum and a grade III splenic rupture (Figs. 1,2). He required transfusion of 5 units of packed red blood cells, 4 units of fresh frozen plasma, and 4 units of cryoprecipitate. The patient underwent splenectomy. His postoperative recovery was uneventful and he was discharged on the sixth postoperative day.

Discussion

To date, rupture of the spleen has been described using many nomenclatures. Liu et al (2019) considered, as did Crate and Payne (1991), that there is a loose application of the term spontaneous rupture. In particular, Liu et al (2019) considered that the term spontaneous could not distinguish a normal spleen from a spleen with pathological changes. For this reason, they suggested the use of the term "atraumatic" because is more accurate and the conditions can be classified according to etiological factors and pathological changes as "atraumatic-pathological splenic rupture" and "atraumatic-idiopathic splenic rupture" (Liu et al, 2019). The incidence rate of ASR was recently determined as 3.2% (Liu et al, 2019). The risk factors for dismal outcomes include splenomegaly, delayed diagnosis, neoplastic

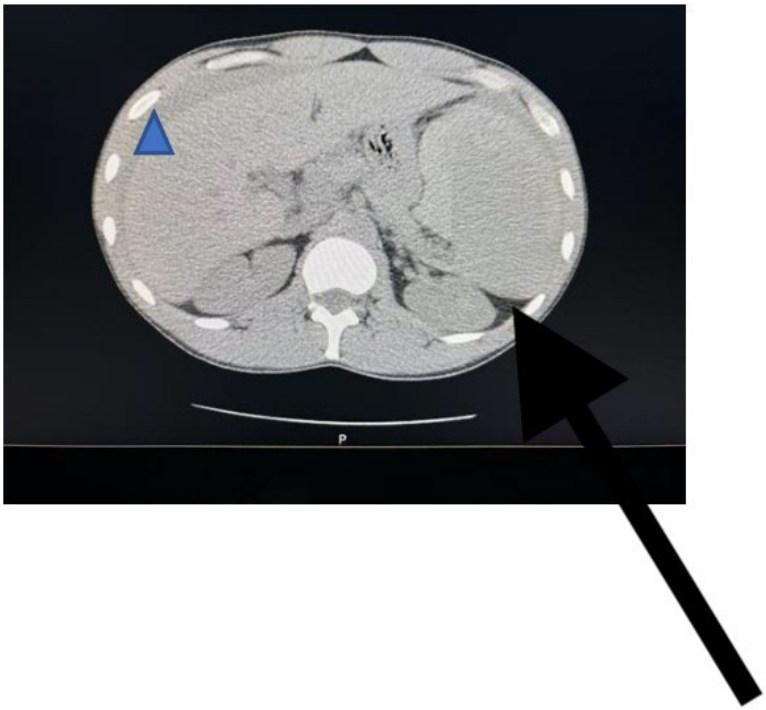

Fig. 1. Computed tomography (CT) scan demonstrating ruptured spleen (long arrow) and haemoperitoneum (short arrow). P, posterior.

disorders, and age over 40 years (Kocael et al, 2014; Liu et al, 2019; Renzulli et al, 2009). In our case with infectious mononucleosis, the reported complication rate of ASR is only 0.1%–0.5% (Farley et al, 1992; Liu et al, 2019).

In particular, in infectious mononucleosis, the spleen appears oedematous, often with subcapsular haematoma. Histologically, the red pulp appears congested with lymphocytic and atypical lymphoid cell infiltration. This infiltration extends to include the supporting trabeculae, blood vessel walls and the capsule of the spleen. As a result, the fibrous support network and the splenic capsule may become dissoluted and fragmented, compromising the spleen's architecture and facilitating rupture (Asgari and Begos, 1997; Farley et al, 1992; Liu et al, 2019).

Of particular interest was the patient was diagnosed by a rural doctor as having acute cholecystitis. Considering that 90% of patients with IM demonstrate hepatitis with mildly elevated serum transaminases in conditions with few diagnostic means, it is easy to misinterpret the symptoms and signs of hepatitis.

In a study of 12 patients with ASR, Kocael et al (2014) demonstrated that the most common etiological factor (33%) was a history of anticoagulant/antiaggregant drug use. Therefore, interactions with other drugs should be considered before prescribing anticoagulants. Caution should be exercised when using anticoagulants in patients with renal or liver dysfunction (Kocael et al, 2014).

When it comes to identifying traumatic splenic injuries, contrast enhanced computed tomography (CECT) has nearly 100% accuracy (Liu et al, 2019; Sortland et

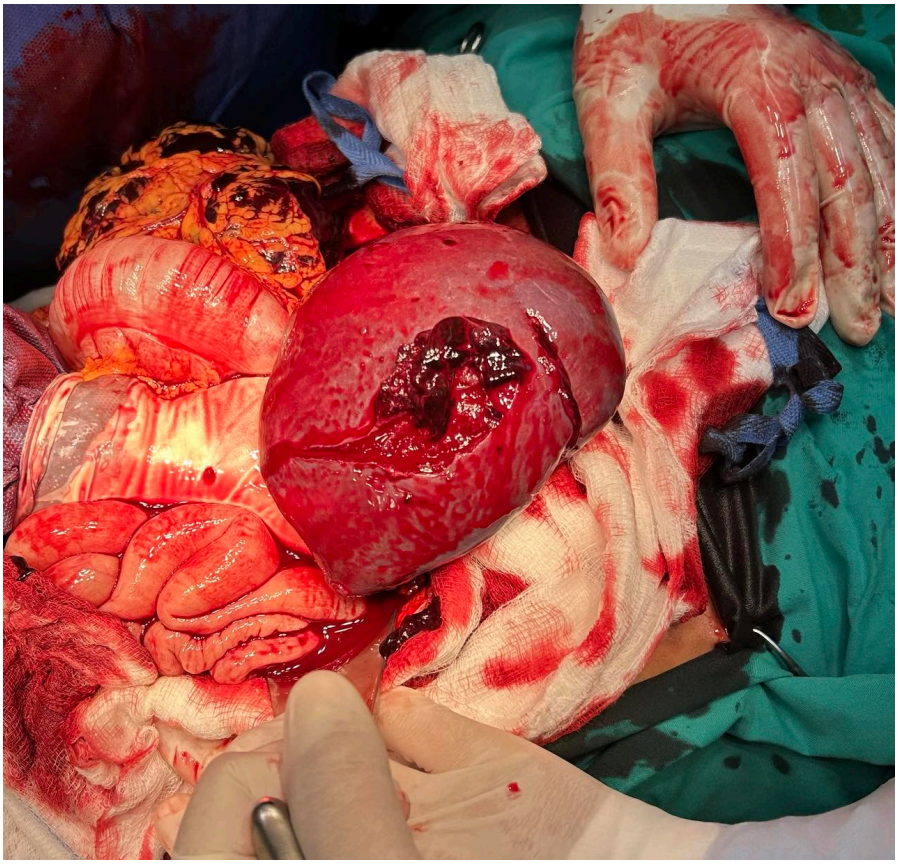

Fig. 2. Operative findings of the enlarged ruptured spleen.

al, 1986). In contrast, the sensitivity of CECT in diagnosing ASR is 85.7% (Kianmanesh et al, 2003; Liu et al, 2019; Sortland et al, 1986). However, the sensitivity of ultrasound for diagnosing traumatic splenic ruptures is 90% (Farley et al, 1992). For the first time, Liu et al (2019) determined the sensitivity of ultrasound for diagnosing ASR and defined it as being at 57.1%. Therefore, CECT is the imaging modality of choice for patients with haemodynamic stability.

According to the American Association for the Surgery of Trauma organ scale injury, most ASRs are grade II or III (Liu et al, 2019).

Treatment strategy decisions were based on haemodynamic status, CT grading, and etiological factors. Conservative treatment can be considered for haemodynamically stable patients with defined etiological factors (Guth et al, 1996; Kianmanesh et al, 2003; Liu et al, 2019; Sortland et al, 1986). However, a high failure rate has been reported (Renzulli et al, 2009).

Total splenectomy is the surgery of choice. This decision is justified for three reasons. First, total splenectomy helps identify underlying pathologies. Second, due to underlying pathologies, the immunological function of the spleen may have been lost. Third, the spleen is oedematous and fragile; therefore, the parenchyma is not adequate to hold sutures (Liu et al, 2019).

Conclusion

Any patient without a history of trauma presenting with left abdominal tenderness, left shoulder tip pain, hypotension, reduced capillary refill time, and low haemoglobin and haematocrit levels should trigger a high index of suspicion to the surgeon regarding an atraumatic splenic rupture.

Learning Points

- In any patient without history of trauma, left shoulder tip pain, low haematocrit and haemoglobin should trigger a high index of suspicion of atraumatic splenic rupture.
- Total splenectomy is the treatment of choice in case of the atraumatic splenic rupture.
- Total splenectomy helps identify underlying pathologies.
- In case of atraumatic splenic rupture the spleen is oedematous and fragile; therefore, the parenchyma is not adequate to hold sutures, so spleen preserved operation is not recommended.

Availability of Data and Materials

All data included in this study are available upon request by contact with the corresponding author.

Author Contributions

PG and PX made substantial contributions to the conception and scope of the manuscript, along with the acquisition and analysis of data. PG and PX were involved in drafting and critically revising the manuscript. Both authors contributed to the important editorial changes in the manuscript. Both authors read and approved the final manuscript. Both authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

The patient consented to his case being published anonymously in the medical literature. The authors declare that the procedures were followed according to the regulations established by the Helsinki Declaration of the World Medical Association.

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Conflict of Interest

The authors declare no conflict of interest.

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