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REVIEW PAPER

Spontaneous spleen rupture in infectious mononucleosis – case report and return-to-play recommendations for paediatric patients

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ABSTRACT

Significant enlargement of the spleen in the course of infectious mononucleosis (IM) may result in spontaneous rupture and life-threatening bleeding. Patients are advised about activity limitation; however, this is based on institution experience and local recommendations. What is important, recommendations for the subgroup of paediatric patients are lacking, and a significant portion of splenic ruptures occur outside the time window of the mentioned current return-to-play recommendations. The aim of this publication was to look closely at the problem of timing and prevention of spleen rupture in the context of IM. Based on clinical experience and retrospective analyses of cases previously published as well as the case report presented, the author proposes a set of recommendations for paediatricians.

KEY WORDS:

children, infectious mononucleosis, return-to-play.

INTRODUCTION

Infectious mononucleosis (IM) is a benign lymphoproliferative disorder caused in 90% of cases by Epstein-Barr virus (EBV) infection. The remaining 10% of IM cases are caused by other viruses such as cytomegalovirus, human herpesvirus 6, toxoplasmosis, HIV, and adenovirus [1]. The classic triad of presenting symptoms consist of pharyngitis, fever, and lymphadenopathy accompanied by others such as malaise, fatigue, and splenohepatomegaly [2]. While childhood infections are usually subclinical, 70% of infected adolescents and adults will develop IM. The Epstein-Barr virus causes lifelong, latent infection by integrating itself into the B lymphocytes, which results in constant low-grade replication and secretion of viral particles to the saliva. It is estimated that 90% of the world's population is infected by adulthood [1, 3, 4]. The diagnosis of IM is based on the serological test – primary infection is characterised by the presence of class M antibodies against viral capsid antigen (anti-VCA IgM) and the absence of antibodies to Epstein-Barr virus nuclear antigen, which appears 2–4 months after the onset of symptoms and persists for the rest of a person's life [5].

The aim of this work is to present a case of a patient with spontaneous spleen rupture in the course of IM and propose a management scheme for patients with IM with respect to "return to play" guidelines.

MATERIAL AND METHODS

After analysing the presented case the authors conducted a search of the literature using a university library search system including databases like PubMed, Chochrane Library, and Scopus for articles published between 1980 and 2022. The keywords entered were spleen

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rupture, infectious mononucleosis, and return-to-play. Of 60 articles found, 4 were selected for further analysis.

CASE REPORT

A 15-year-old patient was admitted to the Department of Paediatric Surgery and Urology due to acute abdominal pain. The pain occurred suddenly when the patient was asleep. There was no known cause of increased abdominal pressure like coughing or sneezing at the time of the spleen rupture. The pain was so severe that it woke the patient and forced him to immediately seek medical help. Additionally, before the onset of the abdominal pain, the patient was suffering from an upper respiratory tract infection with predominant malaise accompanied by sore throat, runny nose, cough, and elevated body temperature (37.8°C) lasting for 4 days. Otherwise, the patient did not have any other acute or chronic condition. At admission to the emergency department, he was in stable condition with heart rate and blood pressure within normal limits. On the physical examination the patient had enlarged cervical lymph nodes predominantly under the angle of the mandible, which was approximately 2–3 cm in diameter. The tonsils were enlarged with a white coating. The abdomen was tender in the epigastric region with slightly expressed peritoneal symptoms. On US the spleen was enlarged to 170 mm with the subcapsular haematoma. The liver was slightly enlarged, and a large amount of fluid in the pelvis was observed. The site of the suspected rupture of the spleen was not visible on US. The imaging studies were extended to abdominal computed tomography (CT), which revealed subcapsular, superficial splenic rupture extending from the middle part to the lower pole with 20-mm thickness subcapsular haematoma. The size of the spleen was 180 mm, and the craniocaudal diameter of the right liver lobe was 180 mm. There was approximately 400 ml of free fluid in the lower pelvis with the radiological appearance of blood. The blood tests showed mild anaemia with the haemoglobin level of 13 g/dl, leucocytosis 13.97 × 10^3/µl with the lymphocytes as predominant cells – 8.39 × 10^3/µl, which accounted for 60.1% of all white blood cells, and normal platelet count of 169 × 10^3/µl. C-reactive protein was 4.1 mg/dl, and the liver enzymes were elevated – alanine transaminase 56 U/l, aspartate transaminase 50 U/l, with elevated total bilirubin level of 1.5 mg/dl and prolonged coagulation time – international normalized ratio (1.33) with normal activated partial thromboplastin time of 35.83 sec. Based on clinical symptoms and laboratory tests, IM was suspected, and a rapid EBV test based on heterophile antibody detection was performed in the emergency department, with positive results. The patient was admitted to the Paediatric Surgery Unit with the diagnosis of spontaneous splenic rupture during the course of IM. The patient remained in a good general condition and was qualified for conservative treatment including intensive fluid administration, strict bed rest, cold compress over the left epigastric region, antibiotic prophylaxis with cefuroxime, and a nil per os regimen. Due to the need for careful fluid management, urine output measurement was started. The patient had the option to choose between bladder catheterization or using a urinal. For the patient's convenience, a 14 Fr Foley catheter was inserted into the bladder and clean urine was obtained. Blood samples for full serological analysis of IM infection were obtained. Due to the altered coagulation status, one unit of fresh frozen plasma was administrated without any side effects. During the first 24 hours the patient was closely monitored – his blood pressure and heart rate were within normal limits. A consecutive blood test was performed, and the results are shown in Table 1. On the first day of observation, oral alimentation was started, with good tolerance. On the 3rd day the results of a serological test for IM were obtained with positive titre of anti-VCA IgM and negative titre of anti-VCA IgG, which confirmed the diagnosis of IM. On the 4th day the abdominal US showed a reduction in the amount of free fluid in the pelvis, to 230 ml, and in the thickness of the subcapsular haematoma, to 12 mm. The antibiotic prophylaxis was ceased. On the 8th day the Foley catheter was removed, and gradual patient mobilisation was initiated, which was complicated by significant abdominal pain. The repeated blood test and abdominal

TABLE 1. Results of the significant blood tests performed during the hospitalisation. Referred values for the blood parameters in our laboratory are shown in square brackets

Day	0	1	2	4	8	11	17
HGB (14–18 g/dl)	13.0/11.6/13.0	10.3/9.9/9.3	9.6/9.9	9.9	11.2	11.6	13.1
HCT (40–54%)	37.6/32.7/36.4	29.2/28.2/26.4	27.3/28	30.2	33	33	35.3
WBC (4–10 × 10^3 µl)	13.97/12.4/7.9	7.5/8.9/8.7	7.38/7.55	6.01	5.59	4.55	5.49
Lymphocytes (24.7–56%)	60.1/–/66.2	59.9/63.3/69	68.6/74.8	76.5	66.4	61.5	59.7
INR (0.9–1.2)	1.33		1.29		1.17		1.19
ALT (10–40 U/l)	56				111		51
AST (15–45 U/l)	50				66		39

ALT – alanine transaminase, AST – aspartate transaminase, HCT – hematopoietic cell transplantation, HGB – haemoglobin, INR – international normalized ratio, WBC – white blood cells

US did not show any signs of active bleeding. The bed rest was reintroduced. On the 11th day gradual patient mobilisation was again initiated, and this time it was well tolerated. On the 16th day the patient was fully mobilised, did not complain of any symptoms, and was discharged from hospital on the 19th day of hospitalisation.

DISCUSSION

Even though IM is a mild disease that affects a substantial percentage of the population and does not raise major clinical concern, it can still become a life-threatening condition. Spleen rupture in the course of IM is a statistically a rare condition with prevalence of 0.1–0.5% [6]. However, when taking into account that approximately 90% of the world's population is going to be infected with the EBV by adulthood, of whom approximately 70% will become symptomatic, the clinical perspective changes substantially.

Spleen rupture in IM is a life-threatening condition with mortality rate of approximately 10% [6, 7]. Although in most of the cases reported as spontaneous (80–86%), this remains controversial [6, 7]. The authors discuss whether the trauma preceding spleen injury was absent or negligible to the patient, knowing that the infiltration by lymphocytes makes the spleen vulnerable to rupture. Even if this is the case and the unobserved trauma can cause splenic rupture, the studies underline the importance of a rest period during the treatment of IM.

The lack of guidelines for the paediatric populations originates in the scarcity of case reports referring to children and adolescent and specific types of publications (reports from sports and military institutes) but the majority of patients described are under 30 years of age [6, 7]. By the age of 30 years, 90% of the worlds' population will be infected with EBV [7]. Knowing that the course of the disease is the same on the histopathological level and that all the reviews incorporate also paediatric patients, the author proposes that the recommendations based on the analysis of the available literature and clinical expertise can also apply to children.

The most widely used recommendations date back to 2008 when the consensus statement of the American Medical Society for Sports Medicine was published [8]. Based on the fact that most of the splenic ruptures take place in the first 3 weeks of illness, the authors suggest that a return to light and contact activity (the difference defined as avoidance of chest and abdominal trauma and significant exertion or Valsalva activities) should be framed for at least 3 weeks after the onset of illness [8]. The authors of more recent publications underline the fact that this recommendation might not be sufficiently anchored in the evidence because the number of splenic rupture cases reported beyond that period remains high [6, 7]. Sylvester *et al.* report that even though most of the cases are reported within the first 3 weeks, 1 in 4 splenic ruptures occurs after the currently recommended 21-day period from the symptom onset. In that cohort 90% of splenic ruptures occurred within 31 days of illness [6]. Overall, the mean time from symptom onset to splenic injury was 15.4 days (SD 13.5 days) and the median time was 11.5 days (range, 1–68 days). The mean time in the group of traumatic patients was 22.9 days (SD 11.9 days) and median of 23 days (range, 1–40 days). The mean time in the group of atraumatic patients was 13.8 days (SD 13.9 days) and the median time was 10 days. Seventeen per cent of ruptures that took place within the first 4 weeks of symptomatic disease suffice to alter the return-to-play recommendations both in the light and contact activity group.

Available demographic data suggest a correlation of young age (mean age 22 years) and sex (70% males) with spleen rupture [7]. This justifies identifying the group of young males as one at highest risk of splenic rupture. Other reports confirm this finding although sometimes the results are biased by the type of publication, i.e. data gathered at military institutes [6].

Abdominal pain is the most common symptom of splenic rupture. With prevalence reaching 90%, it is followed by referred left shoulder pain due to diaphragmatic irritation by bleeding known as a Kehr sign (33%) and collapse or circulatory shock (27%). For the assessment of bleeding in children a drop in haematocrit may give more reliable information of active bleeding than a decrease in blood pressure. None of the ruptures were reported as asymptomatic [7].

The management of patients with spleen rupture remains heterogenous. Bartlett *et al.* report that successful non-operative management was achieved in 28% of cases. Among non-operative strategies are embolization, fibrin tissue adhesive, and serial radiologic control [7]. Other report quotes 90% of successful non-operative-management of patients of which 14.3% were observed in the intensive care setting [6]. One might expect that, with the evolution of solid organ rupture management towards a less aggressive approach, the latter will better describe the future trend.

Splenomegaly as a result of lymphocytic infiltration correlates with the risk of injury as the stretched splenic capsule and increased bulk of the splenic pulp weaken the structure of the organ [9]. Studies assessing the prevalence of splenomegaly in abdominal ultrasounds (US) show that all IM patients present splenomegaly when assessed by US [10, 11]. The spleen reaches its largest size around the 3rd week of disease (23 days), and by the end of week 8 its size normalizes [10, 12]. Spleen size as the only indicator of the return-to-play guidelines is difficult to use because the baseline dimensions are rarely available. However, with serial US, especially in the group of patients with low compliance and greater pressure to quickly return to normal physical activity, decreasing spleen size might be a useful guide.

Blood parameters when assessed were found to be deranged in 90% of cases; however, the size of the spleen

does not correlate with blood parameters, and they are not routinely checked [7, 11]. Also considering that the diagnosis of IM is often established after the spleen rupture, transaminase levels do not seem to serve as a useful marker in return-to-play guidelines.

The outcome in most cases is favourable; however, the mortality rate reaches 9%, with all the fatal cases occurring within the first 2 weeks of disease (0–10) [7]. Remembering that onset of symptoms is often insidious and unspecific and the timing of rupture is not always accurate, it makes the initial weeks of rest very importance. Mild symptoms do not equate to low risk of splenic rupture [6].

CONCLUSIONS

The radiological modality of choice for the exclusion of spleen rupture is angio-CT. A rest period and avoidance of physical activity during IM is advised, in order to avoid spontaneous or trauma-related spleen rupture.

Significant enlargement of the spleen in the course of IM may result in spontaneous rupture and life-threatening bleeding. Patients are advised to limitat activity; however, this is based on institution experience and local recommendations. Recommendations for the subgroup of paediatric patients are lacking, and a significant portion of splenic ruptures occur outside the window of the mentioned current return-to-play recommendations.

The aim of this publication was to look closely at the problem of timing and prevention of spleen rupture in the context of IM. Based on clinical experience and retrospective analyses of previously published cases, the author proposes a set of recommendations for paediatricians.

The group at highest risk of splenic rupture are male patients within the first 4 weeks from the onset of symptoms.

Mild symptoms of IM do not equate to low risk of splenic rupture.

Patients should be asymptomatic when returning to physical activity.

Light activities (without risk of chest or abdominal trauma, avoidance of Valsalva manoeuvres) may be resumed 4 weeks after the onset of symptoms.

Strenuous physical activities may be resumed 8 weeks after the onset of symptoms.

In the case of earlier return to strenuous physical activities and before returning to high-risk sports, radiologic assessment (preferably ultrasound examination) of splenic size should be performed.

Patients should be recommended immediate attendance to the emergency department if they experience abdominal pain, left shoulder pain, light-headedness, or collapse.

DISCLOSURE

The authors declare no conflicts of interest.

REFERENCES

1. Lennon P, Crotty M, Fenton JE. Infectious mononucleosis. BMJ 2015; 350: h1825.
2. Luzuriaga K, Sullivan JL. Infectious mononucleosis. N Engl J Med 2010; 363: 1486.
3. Babcock GJ, Decker LL, Volk M, Thorley-Lawson DA. EBV persistence in memory B cells in vivo. Immunity 1998; 9: 395-404.
4. Thorley-Lawson DA, Miyashita EM, Khan G. Epstein-Barr virus and the B cell: that's all it takes. Trends Microbiol 1996; 4: 204-208.
5. Centers for Disease Control and Prevention. Epstein-Barr virus and infectious mononucleosis, laboratory testing. www.cdc.gov/epstein-barr/laboratory-testing.html.
6. Sylvester JE, Buchanan BK, Paradise SL, Yauger JJ, Beutler AI. Association of splenic rupture and infectious mononucleosis: a retrospective analysis and review of return-to-play recommendations. Sports Health 2019; 11: 543-549.
7. Bartlett A, Williams R, Hilton M. Splenic rupture in infectious mononucleosis: a systematic review of published case reports. Injury 2016; 47: 531-538.
8. Putukian M, O'Connor FF, Stricker P, et al. Mononucleosis and athletic participation: an evidence-based subject review. Clin J Sport Med 2008;18: 309-315.
9. Siliezar MM, Munoz CC, Solano-Iturri JD, et al. Spontaneously ruptured spleen samples in patients with infectious mononucleosis. Analysis of histology and lymphoid subpopulations. Am J Clin Pathol 2018; 150: 310-317.
10. Hosey RG, Kriss V, Uhl TL, DiFiori J, et. al. Ultrasonographic evaluation of splenic enlargement in athletes with acute infectious mononucleosis. Br J Sports Med 2008; 42: 974-977.
11. Dommerby H, Stangerup SE, Stangerup M, et al. Hepatosplenomegaly in infectious mononucleosis, assessed by ustrasonic scanning. J Laryngol Otol 1986; 100: 573-579.
12. O'Connor TE, Skinner LJ, Kiely P, et al. Return to contact sports following infectious mononucleosis: the role of serial ultrasonography. Ear Nose Throat J 2011; 90: E21-24.