Atraumatic Rupture Of A Splenunculus 30 Years Following Splenectomy For Idiopathic Thrombocytopenic Purpura: A Case Report

K Hardiment, J Gunn

Citation

Abstract
We report the unique case of an atraumatic splenunculus rupture in a patient 30 years following elective splenectomy for idiopathic thrombocytopenic purpura and discuss the behaviour and pathology associated with splenosis. We report the case of atraumatic rupture of a Splenunculus occurring thirty years following splenectomy for idiopathic thrombocytopenic purpura.

CASE REPORT
A 40 year old man presented to accident and emergency with acute abdominal pain, which woke him from sleep. At age 10, he had undergone a splenectomy for idiopathic thrombocytopenic purpura. He was not taking any medication and was otherwise fit and well. Initially he was haemodynamically stable, but was pale and sweaty with tenderness and guarding in the left upper quadrant. His blood results were normal other than a mild leucocytosis. A plain abdominal film showed small bowel loops that were shifted towards the right upper quadrant (Figure 1).

Figure 1
Figure 1: Plain abdominal film showing shifted small bowel loops.

An urgent CT scan was performed which showed a large heterogeneous mass lesion in the left anterior pararenal space with free fluid within the peritoneal space consistent with blood (Figure 2). Delayed imaging showed the fluid levels to become more exaggerated confirming active
bleeding (Figure 3).

**Figure 2**
Figure 2: CT Abdomen showing heterogeneous mass lesion with free fluid within the peritoneal cavity

An emergency laparotomy was performed revealing 4 litres of blood in the abdomen and a solitary splenunculus, lying free within the lesser sac. The Splenunculus was extracted and its blood supply, which appeared to arise from the splenic artery, was controlled. The patient required a 6 unit blood transfusion and post-operatively did well. He made a full recovery and was commenced on a re-vaccination programme before being discharged home.

Histopathology showed a splenunculus with appearances consistent with, although not definitive of, idiopathic thrombocytopenic purpura.

**DISCUSSION**

Splenosis can be defined as splenic tissue occurring at an ectopic site as a result of implantation and subsequent growth of original splenic tissue. Numerous reports have documented the existence of splenosis following splenectomy for traumatic rupture and to a lesser extent following splenectomy for medical illness. Splenoses can occur throughout the peritoneal cavity, the thorax and even at distant sites. They tend to be multiple, nodular and usually asymptomatic. Rare complications such as haemorrhage and intestinal obstruction have however been reported. It has been postulated that residual splenic tissue may confer a degree of immunity reducing the likelihood of sepsis following splenectomy for trauma.

Atraumatic rupture of the spleen occurs rarely and is termed pathological if as a result of an underlying disease process, such as malaria or glandular fever. Rupture is termed spontaneous if no such cause can be found. Atraumatic rupture of splenosis is very rare, our literature search yielding just five reports (3, 7, 8, 9, 10). Of these, four appeared to be entirely spontaneous with one occurring possibly due to glandular fever. In all cases however, the original splenectomy was performed due to trauma. Our case therefore is original in that the original splenectomy was performed for medical illness.

It is difficult to draw any firm conclusions regarding splenotic rupture from such a small number of reports. Whereas atraumatic rupture of the normal spleen tends to have a pathological cause, this may not be the case with splenosis as we have no conclusive evidence to suggest that it behaves similarly to its parent tissue. It seems that there is a latent period of several years between splenectomy and rupture, which could possibly indicate that as the lesions grow, their blood supply becomes friable. This may be why, in multiple splenotic nodules; it tends to be the largest which haemorrhages.

Idiopathic thrombocytopenic purpura has been documented to result in atraumatic rupture of the spleen and so may have been a causative factor in our case of ruptured splenunculus. Another possible cause could be an infection, such as glandular fever, which might account for the patient's raised white cell count. Investigation into this cause however was superfluous to the initial management and was therefore not done.

In conclusion, splenosis is generally considered a benign and possibly beneficial condition. Splenosis can however be
occasionally harmful and sometimes result in life-threatening pathology. It is useful therefore to bear in mind the possibility of splenosis and its complications when encountering a previously splenectomised patient with abdominal pain.

CORRESPONDENCE TO
Miss Katherine Hardiment 10 Bigby High Road BRIGG North Lincolnshire DN20 9EZ, UK Telephone: 07787182082 Email: katiekipkins@yahoo.com

References
Author Information

Katherine Hardiment, M.R.C.S.
Department of Academic Surgery, Castle Hill Hospital

James Gunn, F.R.C.S.
Department of Academic Surgery, Castle Hill Hospital