



On-line Case Report

Diagnostic splenectomy for visceral leishmaniasis

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ABSTRACT

A 57-year-old-man with a history of malaise, fever, night sweats and shortness of breath presented a diagnostic challenge to his medical team. He was pancytopenic and had splenomegaly on admission but other investigations, including bone marrow aspiration, proved inconclusive. After the patient deteriorated clinically, the general surgical team was requested to perform a diagnostic splenectomy. The histology of this showed infection with visceral leishmaniasis. He recovered completely with Amphotericin treatment. Although this is a rare condition, particularly for the general surgeon, this case highlights the difficult position surgeons are often put in when performing major surgery diagnostically.

Keywords: Splenectomy – Visceral leishmaniasis – Lymphoproliferative disease – Splenomegaly

Case report

A 57-year-old man presented with a 10-day history of increasing malaise with associated fever, night sweats, epigastric pain, weight loss and shortness of breath. His only notable past medical history was a 7-year history of rheumatoid arthritis treated with weekly methotrexate. He also had a holiday home in Southern Spain. On admission to the gastroenterology ward, his baseline observations were normal although he was markedly pale. Further examination was unremarkable apart from some tenderness and guarding in the left upper quadrant. He had no palpable lymphadenopathy. Blood tests revealed pancytopenia, raised ferritin and IgG. Cultures, chest X-ray, urinalysis (including Bence Jones protein), and myeloma screen, however, were all normal. Computed tomography scanning of the abdomen and thorax revealed a 16-cm spleen with para-aortic lymphadenopathy. Bone marrow aspiration was inconclusive but did not rule out a lymphoproliferative disease. At 10

days post admission, the patient was still spiking temperatures, his shortness of breath increased further and he began bleeding significantly rectally, requiring a transfusion of 4 units of blood.

Considering this clinical deterioration and diagnostic difficulty of his case (he had no accessible lymph nodes), his clinical team thought the best method of securing a diagnosis would be by performing a splenectomy. The general surgical team was consulted and asked to remove the spleen; they agreed. This was performed via a subcostal incision (the procedure was started laparoscopically but proved too technically difficult as the spleen was very friable). The patient's clinical condition improved rapidly postoperatively. Subsequent histology of his spleen revealed infection with visceral leishmaniasis. Interestingly, a retrospective look at the bone marrow still did not reveal any parasites. Following advice from the London Hospital of Tropical Diseases, he commenced treatment with daily Amphotericin B 4 mg/kg weekly in addition to the standard post-

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splenectomy therapy of long-term prophylactic antibiotics, HIB and Pneumonvax vaccinations. He recovered completely from the infection and operation and was able to go home 20 days' postoperatively.

Discussion

Visceral leishmaniasis (also known as kala-azar and black fever) is the most severe form of a disease caused by the sand fly-borne parasite *Leishmania donovani*. Although there is an estimated incidence of 500 cases world-wide, there have only been 47 reported cases in the UK in the last 20 years.¹ The parasite migrates to lymphatic organs, invading macrophages and causing symptoms of fever, weight loss, anaemia and substantial hepatosplenomegaly, often confused (as in our case) with lymphoproliferative malignancy. Misdiagnosis is extremely dangerous as increased susceptibility to opportunistic infections results in a mortality rate of close to 100% if the underlying leishmaniasis is left untreated. Diagnosis is usually made by directly visualising parasites in a bone marrow aspirate. Splenic aspiration is more sensitive but carries a 1% mortality,² which is higher than most general surgeons would expect for a splenectomy, and was the reason for the choice of procedure in this case. Serological testing using the K39 dipstick test is simple, accurate and frequently used in areas where the disease is endemic.³ Other, more sensitive, serological tests currently under development include the use of ELISA, direct agglutination and the Leishmaniasis Skin Test.⁴ Optimal treatment should be commenced promptly. Currently preferred therapy is Amphotericin as described in the

case report; other therapies have been shown to cause unacceptable side-effects and development of resistance in the parasite.⁵

Conclusions

This case highlights two important issues. Although a rare presentation to the general surgeon tropical diseases are becoming more common as exotic travel is extended, in this case only as far as Spain. In addition, it demonstrates the often difficult position a surgeon can be placed in by his physician colleagues when performing major surgery as a diagnostic tool. Although the splenectomy found the diagnosis, one wonders whether repeated bone marrow aspirations may have elucidated the diagnosis and prevented the risks of major surgery and the inherent morbidity.

References

1. Malik A, John L, Bryceson A, Lockwood D. Changing pattern of visceral leishmaniasis, United Kingdom, 1985–2004. *Emerg Infect Dis* 2006; **12**: 1257–9.
2. Da Dilva MR, Stewart JM, Costa CH. Sensitivity of bone marrow aspirates in the diagnosis of visceral leishmaniasis. *Am J Trop Med Hygiene* 2005; **72**: 811–4.
3. Chappuis F, Rijal S, Soto A, Menten J, Boelaert M. A meta-analysis of the diagnostic performance of the direct agglutination test and rK39 dipstick for visceral leishmaniasis. *BMJ* 2006; **333**: 723–6.
4. Singh S, Sivakumar R. Recent advances in the diagnosis of leishmaniasis. *J Postgrad Med* 2003; **49**: 55–60.
5. Sundar S, Chakravarty J, Rai V. Amphotericin B treatment for Indian visceral leishmaniasis: response to 15 daily versus alternate-day infusions. *Clin Infect Dis* 2007; **45**: 556–61.