

Primary research

Spontaneous rupture of malarial spleen: two case reports and review of literature

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Abstract

Malaria has long been among the most common diseases in the southeast Anatolia region of Turkey. In 1992, 18 676 cases were diagnosed in Turkey, and Diyarbakir city had the highest incidence (4168 cases), followed by SanliUrfa city (3578 cases). Malaria was especially common during 1994 and 1995, with 84 345 and 82 094 cases being diagnosed in these years, respectively. Spontaneous rupture of malarial spleen is rare. We saw two cases during 1998, which are reported herein. Both patients were male, and were receiving chloroquine treatment for an acute attack of malaria. One of the patients had developed abdominal pain and palpitations, followed by fainting. The other patient had abdominal pain and fever. Explorative laparotomy revealed an enlarged spleen in both patients. Splenectomy was performed in both patients. We have identified 15 episodes of spontaneous rupture of the spleen in the English language literature published since 1961. Because of increased travel to endemic areas and resistance to antimalarial drugs, malaria is a major medical problem that is becoming increasingly important to surgeons worldwide. Malaria is a particularly important problem in the southeast Anatolia region of Turkey. Prophylactic precautions should be taken by tourists who travel to this region, especially during the summer.

Keywords: malaria, prophylactic precautions, ruptured spleen

Introduction

Involvement of the spleen in malaria that results in splenomegaly renders this organ prone to complications such as rupture [1]. In areas where malaria is endemic, spontaneous rupture of spleen is uncommon. Because of increased travel to endemic areas and resistance to anti-malarial drugs, however, malaria is a major medical problem, which is becoming increasingly important to surgeons worldwide. During the 1980s and 1990s in the USA, the annual number of diagnosed cases of malaria rose by twofold to fourfold that of the preceding decade [2,3]. We report two cases of spontaneous rupture of malarial spleen treated with splenectomy.

Case reports

Case 1

A 56-year-old man presented at hospital with sudden onset of upper abdominal pain, followed by palpitations and fainting. He had been taking chloroquine. In the emergency department he was hypotensive and had pain in the left hypochondrium. Ultrasound showed an enlarged, ruptured spleen and free blood in the abdomen. There was free blood (1500 cm²) in the peritoneal cavity, and a friable, enlarged, ruptured spleen (grade III) was found and removed. Pathological examination revealed that the spleen weighed 950 g and measured 21 × 12 × 7 cm. Gross examination showed the spleen to be dark grey,

Table 1**Reported cases of malaria in Turkey from 1992 to 1998**

| Month | Years | | | | | | |
|-----------|-------|-------|-------|-------|-------|-------|-------|
| | 1992 | 1993 | 1994 | 1995 | 1996 | 1997 | 1998 |
| January | 90 | 82 | 570 | 1032 | 973 | 413 | 194 |
| February | 80 | 96 | 335 | 1496 | 1456 | 732 | 492 |
| March | 204 | 253 | 1132 | 3509 | 3328 | 1375 | 998 |
| April | 565 | 1002 | 2942 | 5081 | 4878 | 1897 | 1367 |
| May | 931 | 1642 | 5557 | 6372 | 8021 | 2996 | 2069 |
| June | 1775 | 3261 | 12745 | 13012 | 8846 | 4304 | 3754 |
| July | 3186 | 6400 | 13700 | 15228 | 10793 | 7342 | 6817 |
| August | 3132 | 8137 | 12033 | 12993 | 7242 | 5890 | 6785 |
| September | 3438 | 9391 | 13221 | 10896 | 6656 | 5069 | 5382 |
| October | 3030 | 9409 | 13285 | 8796 | 5746 | 3383 | 5045 |
| November | 1812 | 6303 | 7006 | 2754 | 2230 | 1533 | 2865 |
| December | 433 | 1234 | 1819 | 927 | 715 | 522 | 1074 |
| Total | 18676 | 47210 | 84345 | 82096 | 60884 | 35456 | 36842 |

with capsular tear and subcapsular haematoma on the medial side. Microscopy revealed dilatation of sinuses, increased pigmentation and elevated concentration of macrophages. Thick and thin films revealed the presence of *Plasmodium vivax*. The patient, much improved, was discharged 6 days after the operation.

Case 2

A 32-year-old man was receiving treatment for a duodenal ulcer in the gastroenterology department. He was also receiving chloroquine for malaria. For 4 days of hospitalization he had high-grade fever and immediately became hypotensive, and had abdominal pain. We performed abdominal paracentesis, which demonstrated free blood in the peritoneal cavity. Explorative laparotomy revealed approximately one litre of blood in the peritoneal cavity and an enlarged spleen, measuring almost double its normal size with a tear on the inferior surface (grade II). Splenectomy was performed. The spleen was 1050 g and measured 22 × 15 × 8 cm. Gross examination showed grayish brown discoloured spleen with capsular tears at a few places. Microscopy revealed congestion and dilatation of sinuses, and bleeding areas. *P vivax* was identified by thick film. The patient was discharged well 11 days after the operation.

Discussion

Malaria parasites have been with us throughout human history. They probably originated in Africa (along with humans), and fossils of mosquitoes show that the

vectors for malaria have existed for at least 30 million years. *Plasmodium* parasites are highly specific, with humans as the only vertebrate host and *Anopheles* mosquitoes as the vectors. This specificity of the parasites also indicates a long and adaptive relationship with humans [4].

Despite initial success, there was complete failure to eradicate malaria in many countries because of a number of factors. Although technical difficulties such as vector and parasite drug resistance have played a part, failure in this endeavour was probably due to social and political factors that prevented efficient application of control measures [4]. The malaria control operation was criticized for being too much like a military operation, and the lack of explanation given to local populations meant that the control measures received little support, or even opposition [5]. Accurate information on the global incidence of malaria is difficult to obtain because reporting is particularly poor in areas that are known to be highly endemic. The global incidence of malaria is estimated to be nearly 120 million clinical cases each year, with nearly 300 million people carrying the parasite. The vast majority of malaria deaths occur in Africa. Estimates vary greatly; the World Health Organization [6] quoted a figure of 800 000 deaths/year in African children during 1991.

Malaria is generally endemic in the tropics, with extensions into subtropic regions. Malaria in travellers who arrive by air is now an important cause of death in nonmalarious

Table 2

Reported cases of spontaneous rupture of malarial spleen since 1960

| Reference | Case no. | Route | Long-term resident of malaria-endemic area | Malarial stage | Species | Symptoms and signs | Surgical therapy | Outcome |
|-----------|----------|---------|--|----------------|---------------------|---|------------------|---------|
| [16] | 1 | Natural | No | Chronic | <i>P malariae</i> | Shock, Kehr's sign LUQ tenderness | Splenectomy | Lived |
| [17] | 2 | Natural | No | Acute | <i>P vivax</i> | LUQ pain, Kehr's sign | Splenectomy | Lived |
| [18] | 3 | Natural | No | Acute | <i>P falciparum</i> | No material | No material | Lived |
| [19] | 4 | Induced | No | Acute | <i>P vivax</i> | Fever, Kehr's sign LUQ tenderness | Splenectomy | Lived |
| | 5 | Induced | No | Acute | <i>P vivax</i> | No material | Splenectomy | Lived |
| | 6 | Induced | No | Acute | <i>P vivax</i> | No material | Splenectomy | Lived |
| | 7 | Induced | No | Acute | <i>P vivax</i> | No material | Splenectomy | Lived |
| [20] | 8 | Natural | No | Acute | <i>P falciparum</i> | Kehr' sign LUQ pain | Splenectomy | Lived |
| [21] | 9 | Natural | No | Acute | <i>P vivax</i> | Fever, back pain severe shock | None | Died |
| [22] | 10 | Natural | No | Acute | <i>P vivax</i> | Fever, hypotension diffuse peritoneal signs | None | Lived |
| [23] | 11 | Natural | No | Acute | <i>P falciparum</i> | Hypotension, Kehr's sign, diffuse peritoneal signs | Splenectomy | Lived |
| [24] | 12 | Natural | No | Acute | <i>P falciparum</i> | Fever, LUQ pain | Splenectomy | Lived |
| [25] | 13 | Natural | Yes | Acute | <i>P vivax</i> | LUQ pain, palpitations | Splenectomy | Lived |
| | 14 | Natural | Yes | Acute | <i>P vivax</i> | LUQ pain, fever | Splenectomy | Lived |
| [26] | 15 | Natural | No | Acute | <i>P malariae</i> | Fever, headache, LUQ pain | None | Lived |
| * | 16 | Natural | Yes | Acute | <i>P vivax</i> | LUQ pain, fever | Splenectomy | Lived |
| * | 17 | Natural | Yes | Acute | <i>P vivax</i> | Fever, palpation, fainting | Splenectomy | Lived |

LUQ, left upper quadrant. *Reported herein.

areas [7], and this is not helped by ignorance or indifference of travellers to prophylactic measures [8].

Malaria has long been among the most common diseases in the southeast Anatolia region of Turkey. During 1992, 18 676 cases were diagnosed in Turkey, and Diyarbakir city had the highest incidence (4168 cases), followed by SanliUrfa city (3578 cases). The southeast Anatolia region is in upper Mesopotamia, covering Diyarbakir, SanliUrfa, GaziAntep, Adiyaman, Batman, Mardin, Siirt and Sirnak. Malaria was especially common during 1994 and 1995, with 84 345 and 82 094 cases being diagnosed in these years, respectively. However, in 1998 the number of new cases decreased, with 36 842 cases being encountered (Table 1). Proquanil with chloroquine have been advised

as the best prophylactic regimen for tourists travelling to the southeast Anatolia region of Turkey between March and November [9].

Spontaneous rupture of the spleen is well described in many diseases, of which malaria is the most common [10,11]. Other such diseases include infectious mononucleosis, splenic neoplasms and haematological malignancies [12–15]. The incidence of rupture of the spleen in malaria is poorly defined. Comparisons of cases that occur in the setting of natural or induced infection have most frequently been employed to investigate this issue. Natural infection is that acquired via a mosquito bite or transplacentally. Induced infection is that obtained through blood transfusion, sharing of needles, laboratory accidents and

experimental or therapeutic inoculation [16]. Fifteen cases of malarial splenic rupture have been reported since 1960 (Table 2). From only three of the reports can the incidence be calculated. Rates of splenic rupture in naturally occurring infection ranged from 0% (0 rupture/5870 total cases of infection) to 2% (1 rupture/51 total cases of infection) [17–19]. The only series of splenic ruptures in induced infections reported an incidence of 0.4% (3 ruptures/715 total cases of infection) [19]. That only 15 cases have been reported since 1960 suggests under-reporting or under-diagnosis, especially for those cases of splenic rupture in the setting of naturally acquired infection [16–26].

Splenic enlargement is very common in malarious areas of the world. It is found in 50–80% of some populations [27]. When a palpable spleen is present, it is generally appreciated within 3–4 days of the onset of symptoms. If disease remains untreated, the spleen can grow. This results in greater average spleen size in a population as the prevalence of malaria increases [15]. *P vivax* has been the species most closely associated with rupture of the spleen in both naturally acquired and induced infections [26]. Among cases reported after 1960, nine out of 15 were due to *P vivax* (four cases were induced infections and five were naturally acquired infections). Both of the patients described above had *P vivax* infection. Spontaneous rupture of the spleen occurs almost exclusively during acute infection, and usually during the primary attack [26].

The exact mechanism of spleen rupture is not known. However, three mechanisms have been implicated in the process [1]. The first of these mechanisms is increase in intrasplenic tension that is due to cellular hyperplasia and engorgement. Second, the spleen may be compressed by the abdominal musculature during physiological activities such as sneezing, coughing, defecation, and sitting up or turning in bed. Finally, vascular occlusion due to reticulate endothelial hyperplasia, resulting in thrombosis and infarction, may be involved. This leads to interstitial and subcapsular haemorrhage and stripping of the capsule, which lead to further subcapsular haemorrhage. The distended capsule finally gives way.

Pathological findings in spontaneous rupture of the spleen include gross and microscopic changes. On gross examination, acutely malarial spleen is dark red because of congestion, hyperaemia and deposition of haemozoin (a malarial pigment). The capsule is thin and friable. Chronic malarial spleen tends to be dark grey, with increased density of connective tissue and fibrosis, resulting in a heavy, often massively enlarged organ with a firm or hard capsule. In addition, disruption of the spleen capsule and single or multiple tears in the underlying parenchyma may be found. Tears may be small or large (grades I–IV) and may be present on any surface. In both of the cases described above, the spleen was massively enlarged.

Microscopic examination demonstrated haemozoin, parasitized and uninfected erythrocytes, and a massive proliferation of macrophages throughout the capillaries, venous sinuses and pulp spaces. There is congestion and dilatation of sinuses and scattered thrombosis, with focal necrosis in capillaries and splenic pulp.

Splenectomy is accepted as the treatment of choice in cases of spontaneous rupture of the spleen. Of the 17 patients described since 1960 (including those described above), 13 patients underwent surgical procedures, all of which were splenectomies. In regions in which malaria is not endemic, improved surgical techniques and supportive care, in addition to increased preoperative and postoperative risk of splenectomy, have led to attempted nonoperative management of splenic rupture in many cases of penetrating and blunt trauma [28]. Nonoperative management consists of observation for 7–14 days in the hospital, strict bed rest, and administration of fluid and blood as needed [25]. In areas in which malaria is endemic, there is growing evidence to suggest that management of spontaneous rupture of malarial spleen without splenectomy should be attempted. A conservative strategy is also reasonable in patients who travel frequently to malarious areas. Splenectomy should be reserved for those patients with severe rupture or those with continued or recurrent bleeding [26].

Conclusion

Changes in the structure of the spleen during the course of malaria can result in asymptomatic enlargement or complications such as haematoma formation and rupture. Primary exposure to malaria and infection with *P vivax* appear to be important factors in spontaneous rupture of the spleen. Malaria is still important in the southeast Anatolia region of Turkey. Prophylactic measures should be taken by tourists who travel to this region, especially those who do so during the summer.

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