Case report

Ruptured hepatic aneurysm in systemic lupus erythematosus

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Summary We report on a patient with systemic lupus erythematosus (SLE) who suffered catastrophic haemorrhage following rupture of an intrahepatic aneurysm. The association between hepatic artery aneurysm and SLE has not been recorded previously, but we have found evidence from the literature that there may be an association between autoaggressive disorders and this surgical emergency.

Aneurysm of the hepatic artery is not common. When intrahepatic, it is associated with systemic disorders. In this report we describe a patient with systemic lupus erythematosus in whom rupture of an intrahepatic artery aneurysm occurred and led to life-threatening haemorrhage.

Case report

An Asian women of 33 presented in 1965 with arthritis and joint swelling. She was anaemic and pyrexial with thinning head hair and pubic hair. A diagnosis of systemic lupus erythematosus (SLE) was made on the basis of marked anaemia, thrombocytopenia, a raised erythrocyte sedimentation rate, raised plasma gammaglobulins, and a positive LE cell test. The patient responded to treatment with 6-mercaptopurine and betamethasone.

Ten years later she was admitted to hospital with arthralgia, pleuritic chest pain, and dependent oedema. X-rays showed bilateral pleural effusions, and an electrocardiogram suggested pericarditis. Further investigations showed antinuclear antibody (positive to 1/50) and raised DNA binding, confirming the diagnosis of SLE. Renal involvement produced proteinuria and diminished function (creatinine clearance 34 ml/min). Frusemide 120 mg with dexamethasone 4 mg was prescribed daily.

Hypertension was treated with bethanidine 60 mg daily.

During the next 2 years her renal function gradually deteriorated, with the creatinine clearance falling to 4 ml/min. In early 1977 the patient was readmitted for further assessment. During the admission she experienced attacks of epigastric pain which were associated with episodes of hypotension. Despite blood transfusion her haemoglobin dropped from 12 g/dl to 3-8 g/dl over 3 days. There was no obvious source of bleeding, but on examination the abdomen was distended by fluid, which diagnostic paracentesis confirmed to be blood. Laparotomy showed the peritoneal cavity to contain 4 l of dark clotted blood, with fresh arterial blood issuing from a small hole on the inferior surface of the left lobe of the liver. An intact subcapsular haematoma occupied the superior aspect of the left lobe of the liver. A left lateral lobectomy of the liver was performed to arrest haemorrhage. Before abdominal closure a renal biopsy was taken.

Postoperatively her renal function continued to deteriorate despite therapy. She died on the eighth postoperative day in renal failure. Permission for necropsy was refused.

The hepatic lobectomy specimen contained a ruptured, fusiform aneurysm, 1 cm in diameter, arising from a branch of the left hepatic artery. Blood had tracked through the otherwise normal liver to form a subcapsular haematoma (Fig. 1). Intraperitoneal haemorrhage followed rupture of the inferior capsule. Renal biopsy showed advanced nephrosclerosis compatible with SLE.
Ruptured hepatic aneurysm in systemic lupus erythematosus

Discussion

Aneurysm of the hepatic artery, though well described, is uncommon. Less than 25% of such aneurysms are said to be intrahepatic (Guida and Moore, 1966), but they have been described in association with systemic infections and pyaemia, trauma, liver abscess, syphilis, and in Marfan’s syndrome. An association between autoaggressive disorders and hepatic artery aneurysm has not been suggested previously. However, reports on such patients incidentally record that 3 were receiving treatment for rheumatoid arthritis (Weaver et al., 1968), 1 for Hashimoto’s thyroiditis (Weaver et al., 1968), 1 for thyrotoxic goitre (Zeppa and Womack, 1963), and 5 for polyarteritis (Glassman and Skerrett, 1960). Autoaggressive disorders appear to account for some 15% of reported intrahepatic artery aneurysms.

There is little doubt that our patient was suffering from SLE as 7 of 14 diagnostic criteria (Cohen et al., 1971) were present. In addition she had antinuclear antibodies and raised DNA binding. An association between SLE and intrahepatic aneurysms does not appear to have been previously described.

Rupture of the aneurysm may not be diagnosed clinically, as the classical features of gastrointestinal bleeding, jaundice, and abdominal pain are present in only one-third of patients (Croom et al., 1976). Without these features the diagnosis is unlikely to be suspected unless suspicion is raised by the recognition of an associated condition. In such cases the diagnosis may be confirmed by selective arteriography. Planned treatment may include lobectomy (Cohen et al., 1966), or, if this is not possible, proximal ligation of the hepatic artery under antibiotic cover should be considered (Erskine, 1973). Superselective arteriography with gel foam embolisation has recently been described and may be a successful method of nonoperative management (Walter et al., 1976).

Further reports are needed to confirm that SLE and other autoaggressive diseases may predispose to the development of this rare but serious surgical emergency.

References

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doi: 10.1136/ard.38.4.396

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