CASE REPORT

Splenic rupture as a consequence of dual malignant pathology: a case report

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A 76 year old woman presented acutely with non-traumatic splenic rupture, which was successfully treated by emergency splenectomy. Histological examination of the spleen revealed the coexistence of metastatic adenocarcinoma cells, together with low grade B cell non-Hodgkin lymphoma. Splenic rupture as a consequence of malignant disease is discussed, together with a brief review of the literature.

Splenic rupture is a rare complication of malignant disease. We present a case report in which the ruptured spleen showed evidence of both non-Hodgkin lymphoma and metastatic breast carcinoma. This acute event was the first manifestation of both malignancies.

CASE REPORT

A 76 year old woman presented with an 18 hour history of epigastric pain and dyspnoea. She had no noticeable comorbidity. On arrival, the patient was hypotensive and tachycardic, with tenderness in the epigastrium. Haematological investigations revealed a normochromic/normocytic anaemia (haemoglobin, 60 g/litre; white blood cell count, $15.8 \times 10^9/\text{litre}$; and platelet count, $88 \times 10^9/\text{litre}$). Renal and liver function tests were within normal limits, as were serum amylase and cardiac enzymes. Ultrasound of the abdomen revealed 17 cm splenomegaly, together with free fluid in the abdomen and pelvis. There was no history of trauma or an obvious cause for the splenomegaly. Laparotomy revealed an enlarged ruptured spleen with several capsular tears and a splenectomy was performed. No other abnormalities at laparotomy were apparent.

The spleen weighed 1295 g and showed severe congestion of the sinusoids, extensive haemorrhage, and splenic vein thrombosis. Scattered throughout the spleen were numerous cells with eccentric nuclei and amphophilic cytoplasm, which stained positive for Alcian blue, indicating that they contained mucin. Immunohistochemistry demonstrated positive staining for polyclonal carcinoembryonic antigen and cytokeratin 7 (CK7), consistent with metastatic adenocarcinoma, but CK20 staining was negative. In addition, there was increased cellularity of the red pulp, and further immunohistochemical studies revealed numerous focal clusters of B cells, which were positive for CD43, focally positive for CD23, and bcl-2, but were negative for CD5, CD10, CD30, CDw66, and cyclin D1. These findings were suggestive of a low grade B cell lymphoma in addition to metastatic adenocarcinoma (fig 1).

A computed tomography scan of the abdomen and pelvis did not identify a primary lesion or lymphadenopathy. The patient made a good recovery from her surgery and was discharged home after 15 days. Four weeks after discharge the patient underwent a bone marrow aspiration and trephine. This showed several B cell lymphoid aggregates with a similar

Figure 1 A double stained section showing large cytokeratin 7 positive (diaminobenzidine, brown) carcinoma cells and small Bcl-2 positive (alkaline phosphatase with fast blue) lymphoma cells in splenic sinusoid.

immunostaining profile to the cells found in the spleen, in keeping with a low grade B cell non-Hodgkin lymphoma. On further review, 10 weeks after discharge, the patient presented with a mobile mass in the lower axilla. This was presumed to be a nodal mass involved with lymphoma and was subsequently excised under general anaesthetic. However, histology showed this to be a 35 mm grade II infiltrating lobular breast carcinoma with vascular invasion. Ten axillary lymph nodes were excised with the mass, none of which showed evidence of metastatic adenocarcinoma or lymphoma. The patient underwent breast radiotherapy and is currently well, taking tamoxifen only.

DISCUSSION

Postmortem studies in patients with cancer show the incidence of splenic metastases to be 2.3–12.9%.¹ Almost all common tumours have been reported at some time to give rise to splenic secondaries, the most frequent being lung, breast, malignant melanoma, and ovary. Berge's large postmortem series found splenic metastases in 7.1% of cases with carcinoma, with the spleen being the 10th most frequent site of metastasis.¹ Splenic secondaries were present in 12% of patients with breast cancer. Splenic metastases are generally seen in patients with advanced disease, with many having metastases at several other sites. Metastatic disease in the spleen is often asymptomatic, but may present with discomfort in the left upper quadrant and symptoms related to pressure on other organs—for example, early satiety and dyspnoea. There may also be features of disseminated systemic disease, such as cachexia and haematological abnormalities as a result of hypersplenism, particularly where there is diffuse parenchymal involvement.

"A recent review of Cancer Registry data found that breast cancer and non-Hodgkin lymphoma occurred together more frequently than expected"

Table 1 Reported cases of splenic rupture secondary to metastatic carcinoma

Primary tumour	Number of cases	Refs
Choriocarcinoma	5	2, 3
Malignant melanoma	4	3
Lung	4	3, 4
Bladder	3	3
Liver	2	3
Teratoma	1	5
Angiosarcoma breast	1	6
Rectal	1	7
Gastric	1	8

Splenic rupture may occur as a result of metastatic disease, although this is rare, with only 22 cases having been reported in the literature to date (Medline, English language).²⁻⁸ Choriocarcinoma, malignant melanoma, and lung cancer account for 59% of these cases (table 1). In the cases where sex was specified, the male to female ratio was 2 : 1, with choriocarcinoma accounting for most of the cases in women. Median age was 48 years (range, 19–84). Although splenic metastases are generally regarded as occurring late in the disease process, splenic rupture occurred at the same time as or before diagnosis of the primary tumour in nine cases. In the remaining cases, the time from diagnosis of the primary tumour to splenic rupture ranged from one month to five years.

Although the spleen is often involved in haematological malignancies, splenic rupture is an infrequent occurrence. In a recent review of the literature, 136 cases of splenic rupture secondary to haematological malignancy were identified.⁹ Acute leukaemia and non-Hodgkin lymphoma were the most frequent, each accounting for 34% of cases, with 18% in chronic myelogenous leukaemia. Male sex, adulthood, severe splenomegaly, and cytoreductive chemotherapy were factors more often associated with splenic rupture.

Synchronous tumours are uncommon and rarely coexist in the same organ. There are several case reports of the coexistence of metastatic breast cancer and non-Hodgkin lymphoma in axillary lymph nodes,¹⁰ but no previous reports of the two occurring together in the spleen. A recent review of Cancer Registry data found that breast cancer and non-Hodgkin lymphoma occurred together more frequently than expected.¹¹ These cases did not appear to be related to previous chemotherapy or radiotherapy, suggesting that the two malignancies may in some cases have a common aetiology. The

Take home messages

- We report splenic rupture as the result of the coexistence of metastatic breast cancer and non-Hodgkin lymphoma in the spleen
- To our knowledge, rupture of a spleen containing two malignant disease processes has not been reported previously.

mouse mammary tumour virus-like ENV gene has recently been suggested as a common link.¹²

Our case is unusual, first in that we report the coexistence of metastatic breast cancer and non-Hodgkin lymphoma in the spleen and, second, because to our knowledge, rupture of a spleen containing two malignant disease processes has not been reported previously.

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