CASE REPORT

Asymptomatic thoracic splenosis after thoracoabdominal trauma: establishing a diagnosis

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Accepted 13 December 2004

Introduction

Splenosis is defined as autotransplantation of viable splenic tissue after splenic injury. Cases of intracranial,\textsuperscript{14} subcutaneous,\textsuperscript{19} pulmonary,\textsuperscript{15} pleural, ovarian, scrotal,\textsuperscript{10} hepatic, pancreatic and peritoneal\textsuperscript{20} splenosis have been reported in the literature after traumatic disruption. Splenosis is common, and when present, typically presents with numerous lesions (up to 100).\textsuperscript{7} The majority of these lesions are found incidentally, masquerading as malignant lesions, but a small number have presented with gastrointestinal or intra-peritoneal bleeding, bowel obstruction,\textsuperscript{1} ureteral colic,\textsuperscript{18} haemoptysis, or neurologic symptoms. The crucial issue with asymptomatic splenosis is to establish a diagnosis of splenosis while excluding malignant lesions in an efficient and minimally invasive fashion. We report a case of multiple pleural splenosis and present it in an historical context.

Case report

A 58-year-old male was referred for evaluation of multiple pulmonary nodules suspicious for malignancy. Thirty-five years earlier, he had been involved in a motor vehicle crash and was diagnosed with a fractured mandible, multiple left-sided rib fractures, a pulmonary laceration with left hemopneumothorax, ruptured left diaphragm with intrathoracic stomach and splenic laceration. After initial assessment and stabilization he underwent splenectomy and repair of the left diaphragmatic laceration. He recovered uneventfully from these injuries and was discharged. Recently, he developed an upper respiratory tract infection and received a chest radiograph (CXR) as part of his investigations. Apical and basal pleural-based nodules were identified CXR (Fig. 1) and further clarified by computed tomography (CT) scan of the chest (Fig. 2). Attempted image guided trans-thoracic aspiration of these lesions in a peripheral hospital was aborted because of fear of complications. Video assisted thoracoscopic surgery (VATS) biopsy of the lesions was performed. Intra-operative frozen section con-
firmed a diagnosis of thoracic splenosis (Fig. 3). A post-operative tagged RBC scan demonstrated two intra-abdominal splenosis in addition to the intra-pleural basilar splenosis (Fig. 4). The patient was discharged on post operative day 2 and was asymptomatic at two subsequent clinic visits.

Discussion

The first case of thoracic splenosis was diagnosed at autopsy in 1937 by Shaw and colleagues, in a 20-year-old male patient who succumbed to overwhelming sepsis. Some time previous he had been in an accident and sustained splenic trauma necessitating removal. Only 17 years earlier von Kuttner described the phenomenon of intra-abdominal autotransplantation of splenic tissue in a single human case and in an experimental canine model. While splenosis within the abdominal cavity is relatively common, occurrence outside the abdominal cavity is unusual. Thoracic splenosis can be suspected if the following factors are present: a history of trauma, splenic injury, and diaphragmatic injury. On reviewing the literature we found 53 previous cases of intrathoracic splenosis in 47 articles. All cases of thoracic splenosis are left sided presumably developing from direct spread of splenic tissue via a diaphragmatic laceration rather than haematogenously. Diaphragmatic lacerations may be subclinical or forgotten at time of presenta-
Of the reported cases, the median age at time of presentation was 45 years with a range of 15–79. All but eight cases were male, reflecting the male predominance in the trauma population. The average time from injury to presentation was 21 years (range 1–49). All lesions were asymptomatic and were found while investigations were performed for angina, vague chest pain, musculoskeletal pain, or when routine imaging was performed. A single exception presented with haemoptysis which resolved upon removal of intrathoracic splenosis.3 Traumatic cases were secondary to sharp or penetrating thoraco-abdominal trauma in 35/54 (64.8%) cases including gunshot and shrapnel wounds and stabblings. A further 13/54 (24.1%) cases were secondary to blunt trauma including falls and motor vehicle accidents and in six cases the cause of trauma was not known.

Diagnosis of thoracic splenosis is difficult and requires the presence of a detailed history as well as a high index of suspicion. Once this diagnosis is entertained it is best confirmed with 99mTc scintigraphy of the chest.4 Both FNA and core biopsy techniques were also frequently employed to establish a diagnosis. FNA is unfortunately often indeterminate as FNA findings in splenosis resemble those of lymphoproliferative disorders.13

When the less aggressive techniques fail, surgery must be undertaken to rule out malignancy with intrathoracic masses. Video assisted thoracic surgery (VATS) is a relatively recent adaptation of minimal access surgery to the thoracic region. Only a single previous report was found which successfully used VATS for biopsy of thoracic splenosis.17 VATS techniques are dependent upon the creation of a working space between the parietal and visceral pleura. Trauma and inflammatory changes can obliterate this potential space with dense adhesions necessitating conversion to conventional open surgery. Thoracotomy was a frequently used diagnostic tool with indication for surgery being the need for histologic confirmation of malignancy.

There are three reports of malignancy concurrent with intra-thoracic splenosis (prevalence 5.6%) and a single report of splenosis presenting as an oesophageal tumor. The first case was a right lower lobe squamous cell carcinoma diagnosed by percutaneous needle biopsy with left multiple pleural nodules proven to be splenosis on 99mTc scanning.2 There were double reports of the next case, a 44-year-old man with a diffuse reticular-nodular pattern and left pleural based mass. Thoracic splenosis was demonstrated with 99mTc scintigraphy and core biopsy, while adenocarcinoma was diagnosed by transbronchial biopsy.6,9 Finally, a right lower lobe T1N0M0 squamous cell carcinoma was misdiagnosed as a T1N3M0 because of “massive left mediastinal lymphadenopathy”.11 The supposed left sided lymphadenopathy was discovered to be benign splenosis on further investigation. A 48-year-old woman presented with a suspected enlarging oesophageal leiomyoma just above the left diaphragm. At the time of thoracotomy, histologic examination identified the suspected leiomyoma as a splenosis.8 These cases highlight the crucial importance of recognizing and differentiating splenosis from primary and secondary neoplastic disease as this affects staging, treatment and prognosis of cancer patients.

Removal of splenosis, while necessary for diagnosis, may be detrimental to a patient. It remains unknown if splenosis can return normal immunological function of the spleen. A recent review reported that up to 92 g of splenosis tissue was insufficient to protect patients from overwhelming post splenectomy sepsis (OPSS).2 On the other hand, studies on immunological function of a patient with previous traumatic splenectomy and 100 intra-abdominal splenosis identified normal circulating antibody levels and response to S. pneumoniae antigens was within normal limits.7 In view of this controversy and potential benefit of splenic nodules we advocate removal of only a minimum of auto-transplanted tissue for diagnosis, rather than excision of all nodules.
The natural history of splenosis in any location is fairly benign. A prospective study of 17 patients using 99mTc imaging demonstrated the risk of developing abdominal and intrathoracic splenosis after combined splenic and diaphragmatic injury as 65 and 27%, respectively.12 There were no reports of enlargement with compression of adjacent structures in the chest causing symptoms. These observations lead us to conclude that routine follow up is not necessary and patients can be discharged to primary care physicians and investigations performed as required for other conditions and risk factors.

Despite confirmation of splenosis with nuclear medicine scanning, concurrent malignancy in other lesions has been reported.2,6,9,11 Investigations must diagnose all lesions suspected of splenosis. When not all lesions can be accounted for, exploration via VATS or thoracotomy is warranted to rule out concurrent malignancy.

Conclusion

Thoracic splenosis remains a rare diagnosis but can be expected based on a history of splenic trauma with diaphragmatic injury and single or multiple left sided pleural based nodules. Diagnosis can be confirmed with tagged RBC scanning and needle biopsy techniques avoiding invasive techniques. When these techniques fail, more aggressive methods are required to distinguish splenosis from malignancy, including VATS or a thoracotomy. Clinical acumen is required to differentiate malignancy from intra-thoracic splenosis, particularly when there are coexisting pathologies in high risk patients. Once diagnosed, splenosis requires neither excision nor frequent follow up.

References