A pleural based mass in a post-partum woman

Padraic C. Ridge [1], Daphne Chen-Maxwell[2], Caroline Brodie[2], Anne Marie Quinn[2], John Bruzzi[3] and David Breen[1]

[1]Interventional Respiratory Unit, Department of Respiratory Medicine, Galway University Hospitals, Galway, Ireland. [2]Pathology Department, Galway University Hospitals, Galway, Ireland. [3]Radiology Department, Galway University Hospitals, Galway, Ireland.

Corresponding author: Padraic C. Ridge (p.macaniomaire1@gmail.com)

Shareable abstract (@ERSpublications)
Can you diagnose this case of a 27-year-old female who presented 1-week post-partum with an incidental finding of intrathoracic masses and probable hilar lymphadenopathy? https://bit.ly/3S3ejVK

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A 27-year-old female smoker was referred to our Rapid Access Lung Clinic from another institution due to an abnormal computed tomography pulmonary angiography (CTPA).

Computed tomography (CT) had been performed on day 1 after an emergency laparoscopic caesarean section due to acute desaturation to 85% on the antenatal ward with associated sudden onset dyspnoea which were transient in nature.

Task 1

Describe the CT findings (figure 1).

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CTPA revealed a left upper lobe pleural based mass with presumed ipsilateral lymphadenopathy extending down to the diaphragm identified on both chest radiography and CTPA (figure 1). There was no evidence of pulmonary embolism or intrapulmonary lesions. With the clearance of the anaesthetic agent and early mobilisation the patient was weaned quickly from supplemental oxygen therapy and subsequently discharged home in good health. This transient oxygen requirement was attributed to post-operative atelectasis, which may have been worsened by a compressive effect from the mass.

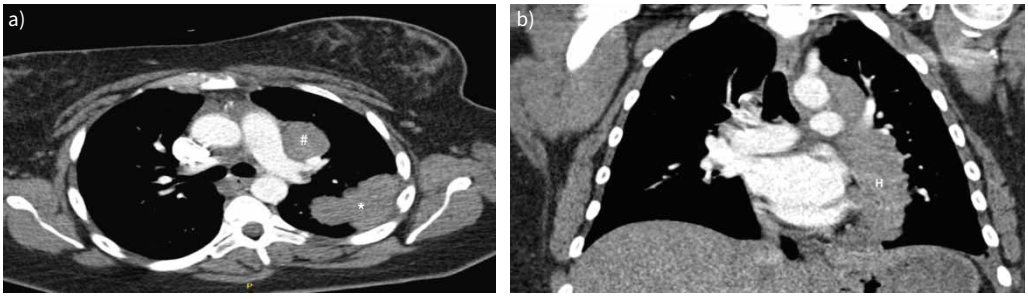

FIGURE 1 a) CT axial view. b) CT coronal view.

She was therefore referred to our rapid access clinic and seen within agreed timelines. She was reviewed 1 week after this event in our clinic and reported no respiratory or systemic symptoms. However, there was a positive family history of lung carcinoma and a personal history of penicillin allergy. She had no known exposure to asbestos or tuberculosis. Cardiorespiratory examination was unremarkable.

Her past medical history was significant for a previous road traffic accident complicated by left hemi-diaphragmatic rupture and splenic injury. This required diaphragmatic repair and splenectomy *via* laparotomy. This had occurred 10 years prior to this presentation.

Routine blood investigations including full blood count, renal profile, liver function tests, C-reactive protein and lactate dehydrogenase (LDH) were normal. There was no prior imaging for comparison.

Task 2

Which of the following are potential diagnoses?
a) Lung cancer
b) Mesothelioma
c) Lymphoma
d) Thoracic splenosis
e) Empyema

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Task 3

What is the most appropriate next investigation?
a) Endobronchial ultrasound transbronchial needle aspiration (EBUS-TBNA)
b) Transthoracic core biopsy
c) Induced sputum
d) Bronchoscopy
e) Surveillance

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We elected to proceed with EBUS-TBNA in our ambulatory interventional respiratory unit and samples were obtained from 11L node. At this point our clinical concern was a diagnosis of lymphoma, lung cancer or thoracic splenosis. We chose EBUS-TBNA as the presumed extensive hilar adenopathy appeared accessible *via* EBUS and it would allow us to stage and diagnose the patient in one procedure, particularly if it confirmed a primary lung malignancy (figure 2). Other factors that influenced this decision were ease of access, lower complication rate and patient choice.

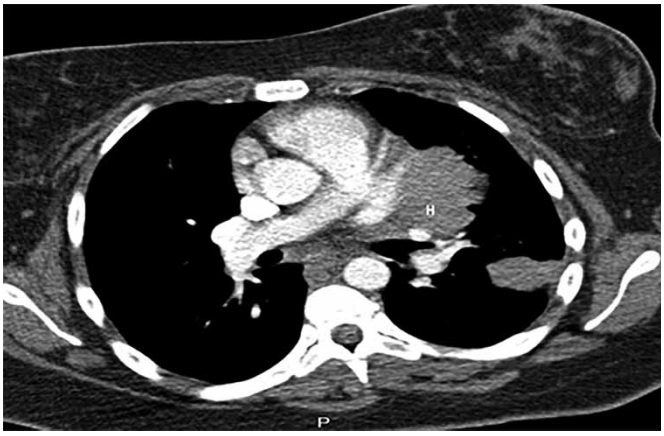

FIGURE 2 CT axial view: hilar abnormality (H) at the orifice of the left lower lobe. Possibly accessible with endobronchial ultrasound transbronchial needle aspiration.

Pathology reported representative material without evidence of malignancy or granulomas. The procedure was poorly tolerated and no other lymph node stations were sampled. In addition, a larger mass that was contiguous with 11L node was identified but unfortunately not sampled.

Task 4

What is the most appropriate next step?

a) Transthoracic needle biopsy
b) Positron emission tomography scan
c) Technetium-99m heat-damaged erythrocyte scintigraphy
d) Repeat EBUS
e) Multidisciplinary meeting

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We discussed this complex case at our multidisciplinary meeting. Due to the hilar involvement, there still remained a clinical concern for a malignant process such as lymphoma. Therefore, a decision was taken to proceed with a transthoracic ultrasound-guided biopsy of the other more peripheral mass. As

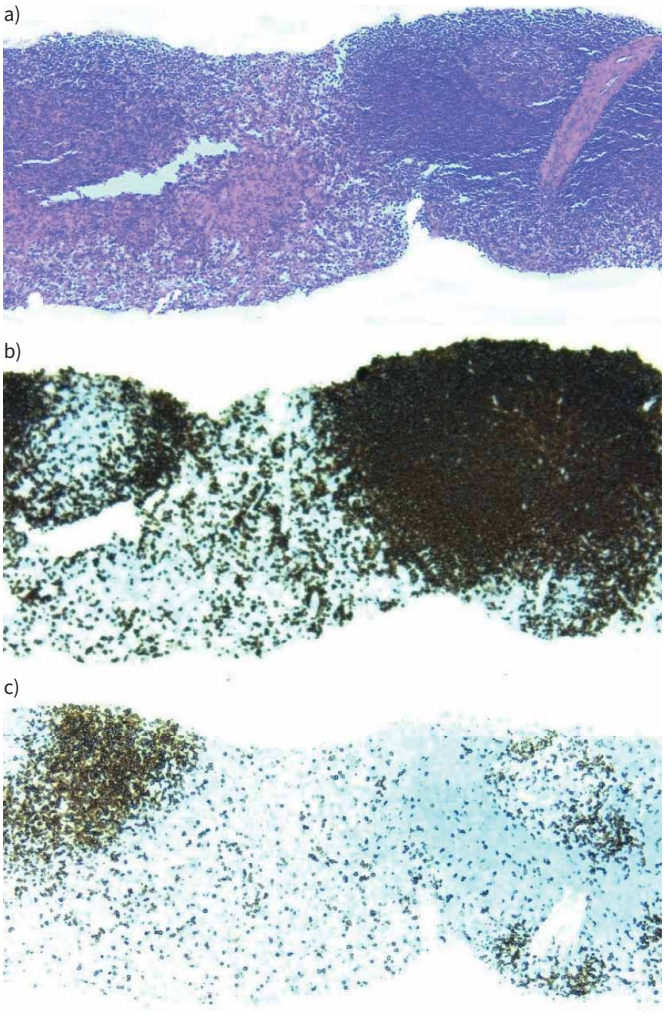

FIGURE 3 a) Haematoxylin and eosin staining (5× magnification) of the core biopsy of left pleura, which shows lymphoid tissue bearing large follicles with central reactive germinal centres and surrounding large marginal zones within background sinusoids. b) Immunohistochemistry (IHC) with CD20 highlights the B-cells within the lymphoid follicles. c) IHC with CD5 highlights the intervening T-cell zones, similar to that seen with CD3. Scale bar=200 μm.

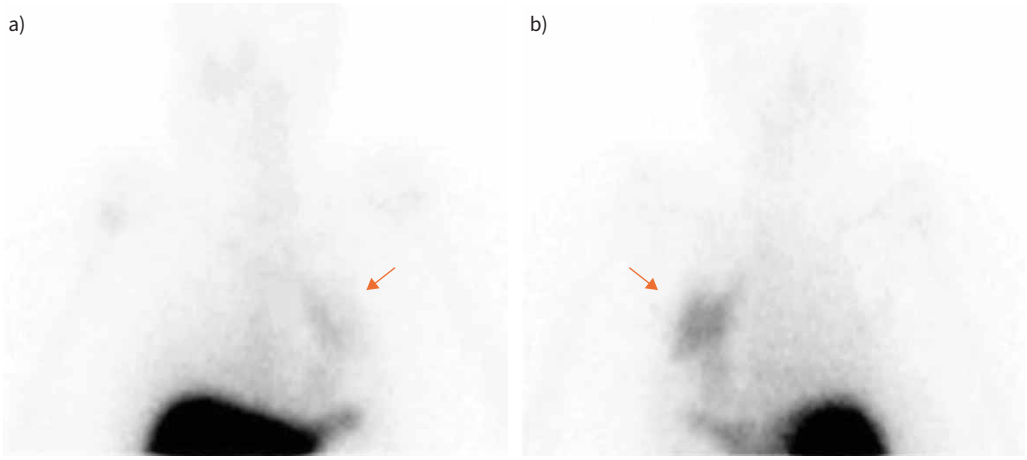

FIGURE 4 Technetium-99m heat-damaged erythrocyte scintigraphy (planar views). a) Anterior and b) posterior views showing erythrocyte uptake in ectopic splenic tissue (arrows). Note the poor anatomical detail based on these images.

this is a core biopsy it would provide more tissue than EBUS-TBNA for the pathologist to examine for the purpose of ruling out lymphoma. In view of the history of previous trauma a nuclear study was recommended as well.

Ultrasound-guided biopsy was performed without complication and histology revealed lymphoid tissue bearing large follicles with central reactive germinal centres and surrounding marginal zones, within background sinusoids. There were no features of high-grade lymphoma. The lesion was felt to be consistent histologically with splenic tissue but a low-grade lymphoma could not be excluded. Given the history of diaphragmatic and splenic trauma, clinically it was felt the material was splenic in origin (figure 3).

A technetium-99m heat-damaged erythrocyte nuclear study was performed and confirmed the pleural based masses were of splenic origin establishing the diagnosis of thoracic splenosis (figure 4).

Task 5

What is the optimal management for this patient?

a) Resection
b) Chemotherapy
c) Embolisation
d) Surveillance
e) Surveillance and antibiotic prophylaxis

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The mass has remained stable on serial chest radiography over 3 years and the patient remains well on erythromycin prophylaxis as recommended by our local antimicrobial guidelines (figure 5).

Discussion

Splenosis is the autotransplantation of splenic tissue post-splenectomy or splenic injury. It is believed that splenic pulp seeds on to the serosal surface where it derives a local blood supply both from bronchial and intercostal sources [4, 9]. It is estimated to occur in the abdominopelvic cavity in up to 65% of cases of splenic rupture. Thoracic splenosis is rarer and is believed to occur only when there is concomitant damage to the diaphragm allowing splenic particles to implant on the parietal/visceral pleura or more rarely in the lung parenchyma [4, 9].

Thoracic splenosis is most often identified incidentally as multiple pleural based masses on radiological imaging [10]. While it is usually asymptomatic it can rarely present with pleuritic pain, cough or haemoptysis [4, 9, 11]. Presentation is often decades after the initial trauma and failure to elucidate the history of splenic injury may result in unnecessary interventions [4].

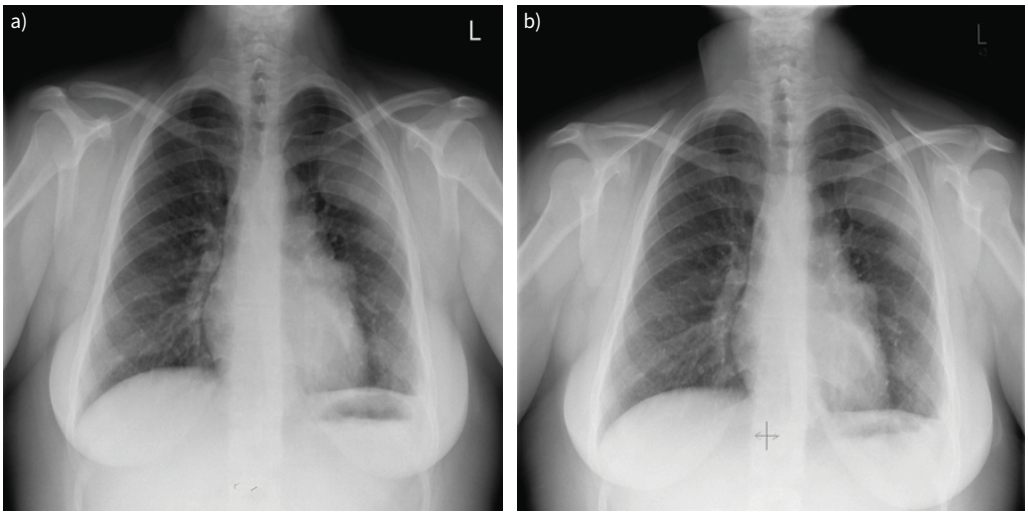

FIGURE 5 Chest radiographs in a) 2016 and b) 2019. Left upper lobe and hilar based masses are unchanged over 3 years of follow-up.

Diagnosis can be made without biopsy using scintigraphy. Nuclear studies using technetium-99m heat-damaged erythrocytes have been shown to be more sensitive and detect smaller deposits in comparison to studies using older agents, such as sulfur colloids. This is a highly specific test as only splenic tissue will sequester and phagocytose the heat-damaged erythrocytes. This allows selective splenic imaging. In isolation, scintigraphy has limitations in specificity with regards to precise localisation of the lesion. As can be seen in our case, the images from scintigraphy do not provide accurate anatomical detail and can be difficult to interpret. The introduction of hybrid imaging using single-photon emission computed tomography (SPECT)-CT has overcome this limitation. Unfortunately, when this case presented to our institution we did not have access to SPECT-CT. Therefore, as our patient had perihilar pleural involvement resembling mediastinal adenopathy, we felt biopsy was required to definitively rule out other mimicking conditions such as lymphoma, mesothelioma and pleural metastasis [11, 12].

For thoracic splenosis management is normally expectant. Deposits of thoracic splenosis derive their blood supply from surrounding tissues. This limited supply restrains their growth over time as manifested over the 3 years of follow-up in our case [7, 13]. Surgery is generally discouraged but may be indicated in those in whom there is a concern regarding malignancy [7, 14]. Surgery has also been performed for control of symptoms such as refractory pain due to local pressure or haemoptysis; however, this has been reported in only a handful of cases [7].

Unfortunately, the ectopic spleen remnants do not provide adequate protection against encapsulated bacteria such as *Streptococcus pneumoniae* (responsible for more than 50% of infections), *Haemophilus influenzae* type b or *Neisseria meningitidis* [8, 15]. These patients are rendered functionally hyposplenic and thus have a 5% lifetime risk of developing overwhelming post-splenectomy infection [15]. Long-term management in those with thoracic splenosis should include standard post-splenectomy care with education, vaccination, antibiotic prophylaxis, malaria prophylaxis for travellers, and rapid management of fevers and animal bites [15].

Conclusion

Thoracic splenosis is a benign pathology and should always be considered in those with a thoracic mass and a history of splenic injury. Failure to illicit this history can result in extensive, invasive investigations and the associated distress and risk of harm to the patient. If resources allow, and in the right clinical context, diagnosis should be attempted noninvasively with nuclear imaging prior to proceeding to invasive procedures.

Key points

- Thoracic splenosis should be considered in those with an intra-thoracic mass and a prior history of diaphragmatic trauma.

- Thoracic splenosis can present as an incidental finding many years after the initial trauma.
- Diagnosis can be made noninvasively with the use of nuclear imaging.
- Management is most often conservative.
- Those with thoracic splenosis are functionally hyposplenic and are at the same risk from encapsulated organisms as patients post-splenectomy.

Answer 1

The CT axial view (figure 1a) shows a large pleural based mass in the left upper lobe (*) and an ipsilateral hilar based mass (#). The CT coronal view (figure 1b) shows a hilar mass extending down to diaphragm (H).

<< Go to Task 1

Answer 2

Options a–d are correct (lung cancer, mesothelioma, lymphoma, thoracic splenosis).

a) Lung cancer: despite the patient's young age, lung cancer is a possibility as the patient is a smoker, has a family history, has an intra-thoracic mass with mediastinal pleural involvement and possible hilar lymphadenopathy [1].
b) Mesothelioma: a pleural based mass with involvement of the mediastinal pleura raises concern for mesothelioma [2].
c) Lymphoma: despite the lack of systemic symptoms or raised LDH, lymphoma is a possibility [3].
d) Thoracic splenosis: in light of the prior history of trauma, diaphragmatic repair and splenectomy this is a possibility. However, her changes are quite extensive and the hilar involvement is atypical [4].
e) Empyema: is unlikely as the patient is systemically well, and has normal blood investigations.

<< Go to Task 2

Answer 3

Option a (EBUS-TBNA) is correct.

a) EBUS-TBNA: this is the most appropriate investigation at this stage as it will aid us to visualise the mediastinum and potentially diagnose and stage the patient in one procedure [5].
b) Transthoracic core biopsy: this test may provide more tissue than EBUS-TBNA. However, if this process turns out to be malignant it will only provide information regarding the T status of the tumour. The patient would need to undergo a second invasive investigation to confirm stage regardless of the outcome of this test. Therefore, the test that provides the highest stage should be the initial test [1].
c) Induced sputum: while this can provide a diagnosis there is no definite evidence of endobronchial disease. This is much less likely than other methods to provide the answer and may delay the investigation pathway [1, 6].
d) Bronchoscopy: there is no definite evidence of endobronchial disease. This test can be done as part of the EBUS procedure but is not the single most suitable test.
e) Surveillance: there are features concerning for malignancy such as mediastinal involvement and hilar lymphadenopathy. This would not be the correct option.

<< Go to Task 3

Answer 4

Option e (multidisciplinary meeting) is correct.

a–d are all potential next steps. They may all add evidence to the final diagnosis. However, this is a complex case and warrants discussion with the multidisciplinary team including a thoracic surgeon, pathologist, interventional respiratory physician and interventional radiologist to decide the best investigation that is most likely to give the diagnosis.

<< Go to Task 4

Answer 5

Option e (surveillance and antibiotic prophylaxis) is correct.

a) Resection: this is incorrect. This would be a large operation with huge morbidity. There is no therapeutic benefit in this asymptomatic patient [7].
b) Chemotherapy: this would not be appropriate as the patient does not have a confirmed malignancy.
c) Embolisation: this patient is asymptomatic so there is no role for this.

d) Surveillance: due to the atypical radiological appearance and the inability to fully rule out a low-grade lymphoma based on histology this would be prudent. Also, we do not know at this point whether the various deposits of ectopic splenic tissue have reached their full size. However, it is not the single most correct answer.
e) Surveillance and antibiotic prophylaxis: in addition to surveillance this patient will need standard post-splenectomy care as we will discuss in further detail [8].

<< Go to Task 5

Conflict of interest: None declared.

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