CASE REPORT

Rare presentation of a localised malignant pleural mesothelioma with cranial metastasis

Gulhan Ertan,[1] Abdulkadir Eren,[1] Sila Ulus[2]

[1]Department of Radiology, Medipol University School of Medicine, Istanbul, Turkey
[2]Acıbadem University School of Medicine, Istanbul, Turkey

Correspondence to
Dr Sila Ulus,
silaulus@hotmail.com

Accepted 19 September 2016

SUMMARY

Mesothelioma is an uncommon malignant neoplasm and a localised form of the pleura is especially very rare. Diagnosis of localised malignant pleural mesothelioma (LMPM) is very challenging. Histopathological verification is the gold standard, and studies such as CT, positron emission tomography (PET) and thoracoscopy are very valuable tools in assisting diagnosis. We report a case of histopathologically proven LMPM, which was discovered as a well circumscribed solitary subpleural nodule on PET-CT after presentation with cranial metastasis. This case shows that LMPM can present with uncommon radiological and clinical appearances, and imaging tools such as PET-CT have a very important role in diagnosis.

BACKGROUND

Malignant mesothelioma (MM) is a rare neoplasm, yet it is the most common primary malignancy of the pleura. It typically presents with pleural effusion or pleural thickening and masses.[1] Most of the mesotheliomas present as a diffuse form, but localised forms are also described. Diffuse form is usually associated with asbestos exposure. Localised malignant pleural mesothelioma (LMPM) is a rare tumour,[2–4] which usually appears as solitary circumscribed nodule or mass attached, either in a sessile manner or pedunculated manner to the surface of the pleura.[5] LMPM should be distinguished from diffuse malignant pleural mesotheliomas, because a good prognosis may be obtained by surgical resection.[2 6] Diagnostic imaging involves: standard X-rays, CT, MRI and positron emission tomography (PET). Combination of chest CT and PET shows a high specificity. The best methods for diagnosis are videothoracoscopy and histopathological verification. We present a case of a male patient with a well circumscribed solitary subpleural nodule discovered on PET-CT scan after presentation with cranial metastasis, who underwent video-assisted thoracic surgery (VATS), which was histopathologically proven to be a LMPM.

CASE PRESENTATION

Three months ago, a 44-year-old male patient was examined because of diplopia and a small parietooccipital cerebral mass detected on MRI at another medical centre. There, he was operated and histopathological examination result was concordant with metastasis. The patient was referred to our clinic for the search of primary malignancy.

INVESTIGATIONS

Initially PET-CT scan, gastroscopy and colonoscopy were performed to rule out lung and gastrointestinal system neoplasms. Gastroscopy and colonoscopy findings were normal. However, PET-CT examination revealed a hypermetabolic subpleural nodule in the left upper lobe lingular segment (figure 1A). Thoracic CT examination showed that the size of the nodule was 15×8 mm and it was oval shaped with smooth contours (figure 1B,C). In addition, emphysematous changes characterised with bullae in the thorax parenchyma more prominent on the upper lobes were noted. The nodule was protruding into a large bulla. There was no pleural thickening or calcification on imaging. The patient had 30 pack-year smoking history. The patient was an accountant and there was no history of asbestos exposure.

DIFFERENTIAL DIAGNOSIS

The lesion appeared highly suspicious and was presumed to be the primary malignancy focus. Differential diagnosis included adenocarcinoma, mesothelioma and benign sequel pleural focal thickening.

TREATMENT

VATS left upper lobectomy was performed because there was very thin parenchyma remaining due to the bulla. Frozen section procedure was carried out during the surgery and tumour within the bulla was detected as a papillary structure. Lymphadenectomy was also performed. No parietal pleural nodule or thickening was seen during VATS.

OUTCOME AND FOLLOW-UP

On histopathological examination, the lesion was diagnosed as LMPM and it had the same immunophenotype with the cranial resection material. No subtype was reported. GATA 3, CK 7 and Kalretinin were positive in the immunohistochemical study confirming the diagnosis. The immunohistochemical expression of thyroid transcription factor-1 was also evaluated and it was negative. Lymphadenectomy dissection results were consistent with reactive lymph nodes with anthracosis. The patient underwent chemotherapy and radiotherapy after the surgery. Three months after the operation diffuse metastases were detected in the brain, liver and bones. Biopsy of the lesions from the liver was consistent with MM metastates. Bilateral pleural effusion and ascites occurred. Radiotherapy was performed for the metastatic brain lesions. The patient died a few months later.

To cite: Ertan G, Eren A, Ulus S. *BMJ Case Rep* Published online: [*please include* Day Month Year] doi:10.1136/bcr-2016-217348

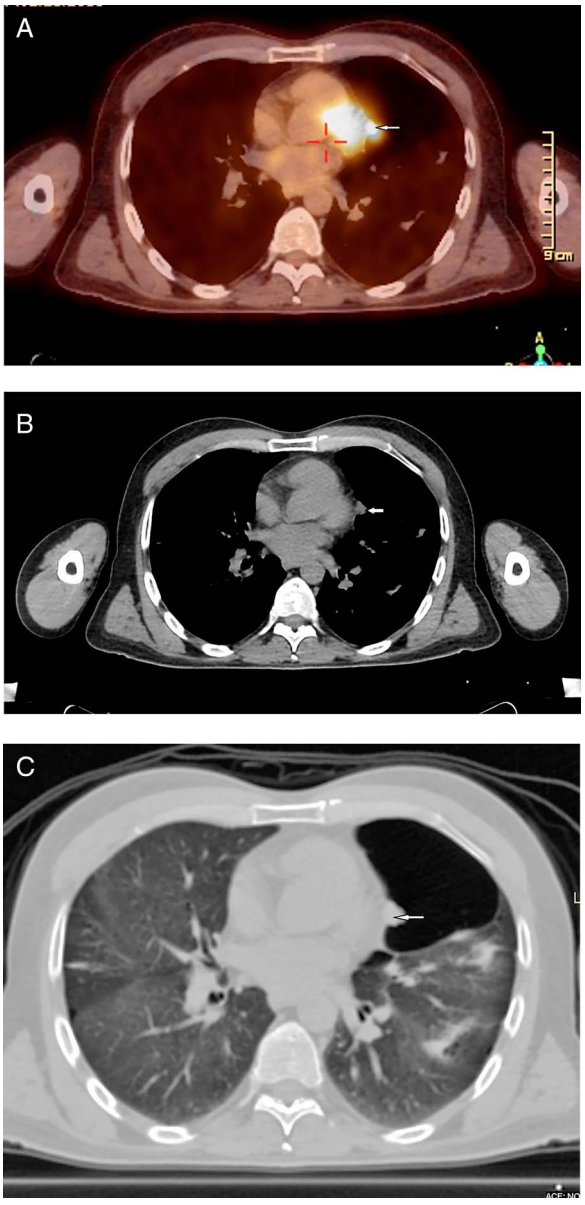

Figure 1 Axial PET-CT image (A) revealing a hypermetabolic subpleural nodule in the left upper lobe lingular segment. The nodule is seen in the mediastinal (B) and parenchymal (C) windows of the thorax CT examination. Note the large bulla adjacent to the nodule in the parenchymal window. PET-CT, positron emission tomography CT.

DISCUSSION

MM is a very aggressive and treatment-resistant tumour making early diagnosis very important.[7] MM usually presents with pleural thickening, calcification and effusion and it is usually associated with long-term asbestos exposure. MM has two forms; diffused and localised. LMPMs are very rare and could present with diffuse parenchymal infiltration without clinically or radiologically detectable pleural lesions.[8] Localised forms are difficult to diagnose.[9] The patients can be asymptomatic or can present with metastasis. In our case, the patient had a solitary subpleural nodule and did not have any pleural thickening or calcification on PET-CT scan. He presented with cranial metastasis, which is identified in about 3% of cases in recent postmortem studies.[10] He also had no history of asbestos exposure. CT scan is the first-line diagnostic tool,[11] but PET-CT scan is the gold standard modality for the evaluation of metastatic disease. In our case PET-CT scan was used as the first step to search for the primary tumour. Since early diagnosis is essential, our case shows the importance of recognising that LMPM can present with uncommon radiological and clinical appearance such as a solitary subpleural nodule or with its metastasis. Imaging tools, especially PET-CT have a very significant role in the establishment of this rare diagnosis.

Learning points

- It should be kept in mind that localised malignant pleural mesothelioma can present with uncommon radiological and clinical appearance such as a solitary subpleural nodule or with its metastasis.
- Imaging tools, especially positron emission tomography CT have a very significant role in the diagnosis of localised malignant pleural mesothelioma.
- Early diagnosis is essential for the treatment of localised malignant pleural mesothelioma.

Contributors GE, AE and SU made substantial contributions to the conception or design of the work, or the acquisition, analysis or interpretation of data. SU and GE contributed by drafting the work or revising it critically for important intellectual content. GE, AE and SU contributed by approving the final version of the manuscript published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1 Tamer Dogan O, Salk I, Tas F, *et al*. Thoracic computed tomography findings in malignant mesothelioma. *Iran J Radiol* 2012;9:209–11.
2 Nakano T, Hamanaka R, Oiwa K, *et al*. Localized malignant pleural mesothelioma. *Gen Thorac Cardiovasc Surg* 2012;60:468–74.
3 Crotty TB, Myers JL, Katzenstein AL, *et al*. Localized malignant mesothelioma. A clinicopathologic and flow cytometric study. *Am J Surg Pathol* 1994;18:357–63.
4 Galateau-Salle F, Churg A, Roggli V, *et al*. World Health Organization Committee for tumors of the Pleura. The 2015 World Health Organization classification of tumors of the Pleura: advances since the 2004 Classification. *J Thorac Oncol* 2016;11:142–54.
5 Haithcock BE, Zagar TM, Zhang L, *et al*. Chapter 73: diseases of the pleura and mediastinum. In: Niederhuber JE, Armitage JO, Dorshow JH, *et al*., eds. *Abeloff' s clinical oncology*. 5th edn. Philadelphia, PA: Elsevier, 2014.
6 Papadopoulos A, Porfyridis I, Christodoulides G, *et al*. A rare clinical case—solitary fibrous tumor of the pleura. *Respir Med Case Rep* 2015;16:117–19.
7 Hassan I, Goolam-Mahomed A. An unusual cause of an anterior mediastinal mass. *Respir Med Case Rep* 2014;12:52–4.
8 Hasegawa M, Sakai F, Sato A, *et al*. FISH analysis of intrapulmonary malignant mesothelioma without a clinically detectable primary pleural lesion: an autopsy case. *Jpn J Clin Oncol* 2014;44:1239–42.
9 Klawiter A, Damaszke T. Pleural mesothelioma—case report. *Pol J Radiol* 2010;75:61–3.
10 Finn RS, Brims FJ, Gandhi A, *et al*. Postmortem findings of malignant pleural mesothelioma: a two-center study of 318 patients. *Chest* 2012;142:1267–73.
11 Yao W, Yang H, Huang G, *et al*. Massive localized malignant pleural mesothelioma (LMPM): manifestations on computed tomography in 6 cases. *Int J Clin Exp Med* 2015;8:18367–74.