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Bilateral pneumothorax in a case of a 20-year-old woman with metastatic malignant mesenchymoma of the pelvis minor and perineum

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Summary

We present a rare case of bilateral, recurrent and persistent pneumothorax, requiring suction drainage, as a complication of metastatic spread of a malignant neoplasm to the lungs.

Key words: malignant mesenchymoma • metastases • chest X-ray • chest CT

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Background

Pneumothorax defined as the presence of air between the pleural layers may occur spontaneously or as a condition secondary to various pathologic or mechanical factors.

The most frequent causes of pneumothorax are thoracic traumas (with fractures of one or more ribs, lung contusion or formation of a bronchopulmonary fistula). Other causes include chronic obstructive lung disease, interstitial disorders (especially the alveolar form of sarcoidosis, LAM, histiocytosis), infections (e.g. with staphylococcus aureus), neoplasms (especially metastases of sarcomas) and iatrogenic ones (therapeutic or diagnostic puncture of the pleura, complications associated with other procedures).

Spontaneous pneumothorax also occurs, most frequently in young, healthy subjects; according to some authors, tall stature may be a predisposing factor.

Among imaging modalities, plain chest radiography (X-ray) in vertical position is the leading one (characterized by high sensitivity and rarely necessitating expiratory imaging). Chest radiography performed in horizontal position is considerably less sensitive.

In case of any doubt, or necessity of extended diagnostics, computed tomography (CT) of the chest is recommended.

Case report

A 20-year-old woman sought medical assistance because of edema of the left lower extremity and a tumor in the left groin.

Preliminary CT of the pelvis visualized an extensive tumor-like mass on the left side in the vicinity of hip acetabulum, causing displacement of the urinary bladder, uterus and rectum to the right. The predominant portion of the mass passed out of the pelvis, surrounding the left pubic bone, the pubic symphysis and the region of the obturator foramen, and descending to the groin. The tumor demonstrated signs of necrosis, contrast enhancement on the periphery, and the pubic bone and pubic symphysis had undergone osteosclerotic remodeling (fig. 1).

Chest X-ray revealed the presence of numerous nodular lesions in the lungs, especially in the left one.

After biopsy of the tumor of the pubic region, which led to the diagnosis of chondroblastic mesenchymoma of the pubic bone, the patient was referred to the Institute of Mother and Child, where ultrasonography (USG) revealed additionally infiltration of the left pudendal lip and medial aspect of the left thigh. Besides displacement of the urinary bladder, infiltration of a ca. 5 cm segment of its wall was also visible (fig. 2a and 2b).

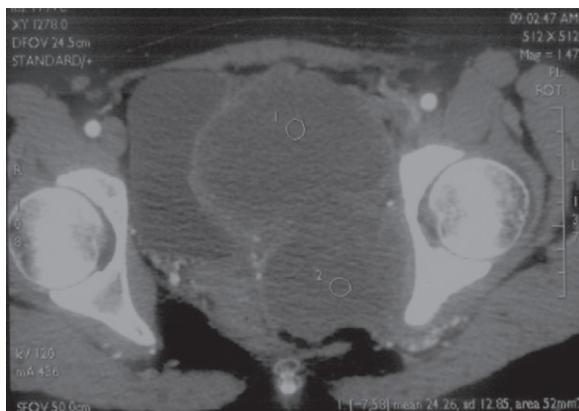


Figure 1. CT, a big tumor in the pelvis minor, inhomogeneous contrast enhancement, with areas of necrosis.

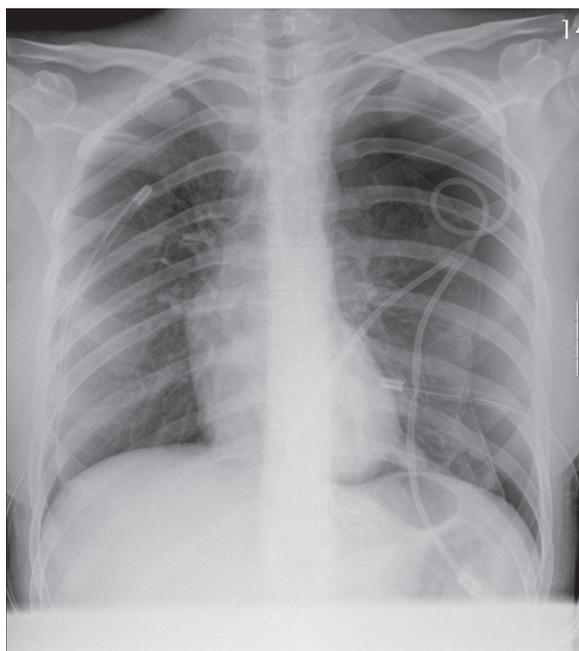


Figure 3. Plain film, bilateral pneumothorax with drainage, discrete metastatic nodules.

Because of persistent vaginal bleeding, a gynecological examination was performed, which revealed neoplastic infiltration of the vagina.

On admission, the patient complained of pain in the left side of the chest during respiration. Chest X-ray revealed left-sided pneumothorax up to 3.5 cm thick and numerous nodular lesions in both lungs, predominantly in the left one. The image suggested metastatic character of the lesions.

Despite drainage, the left-sided pneumothorax persisted during hospitalization, periodically increasing to 5 cm. After 10 days of hospitalization, up to 5.4 cm right-sided pneumothorax appeared, which also required drainage (fig. 3).

Scintigraphy demonstrated accumulation of radioisotope in the region of the left hip joint, adjacent to the ischial and pubic bone, as well as extraosseous uptake in the superior and medial part of the left thigh.

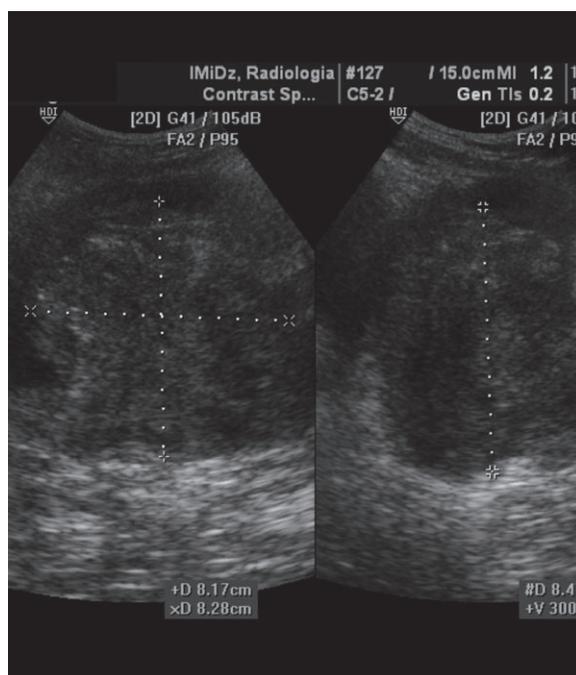


Figure 2 A. US, a large cystic-solid mass in the pelvis, with irregular necrotic areas.

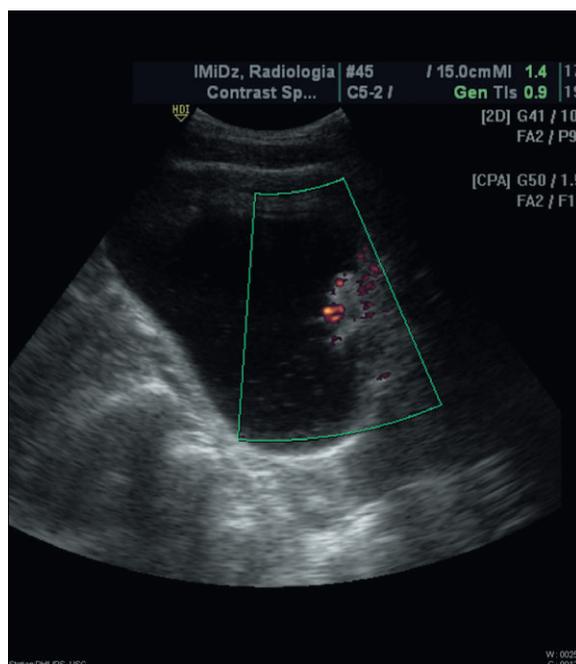


Figure 2 B. US, urinary bladder wall infiltration.

Because of clinical doubts, the tumor biopsy was repeated, and on that basis the most probable diagnosis of mesenchymoma malignum of mixed texture, mainly chondro-muscular was established.

Despite suction drainage and chemotherapy, the bilateral pneumothorax persisted. Its presence, with left-sided thickness up to 2.8 cm, and right-sided up to 1.6 cm, was confirmed by chest CT, which also revealed numerous, bilateral, thick-walled lesions containing air, both peripheral and

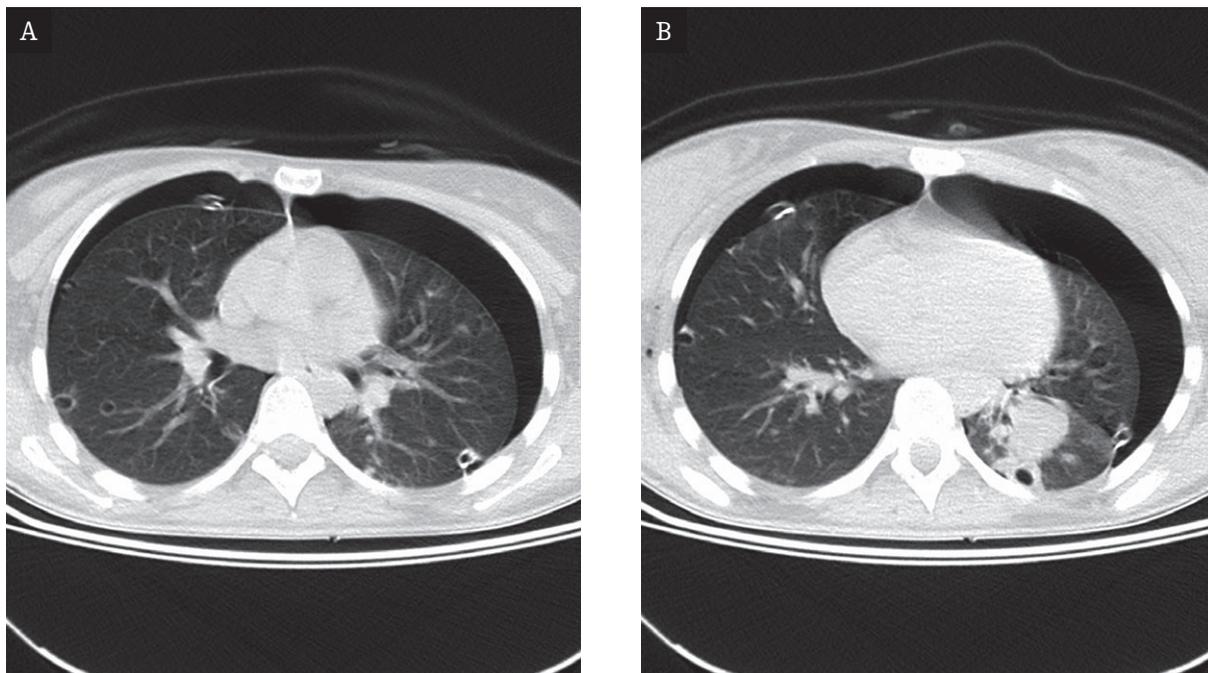


Figure 4 A, B. CT, cystic and solid changes, some of them localized at the margins of the lung tissue. Bilateral pneumothorax.

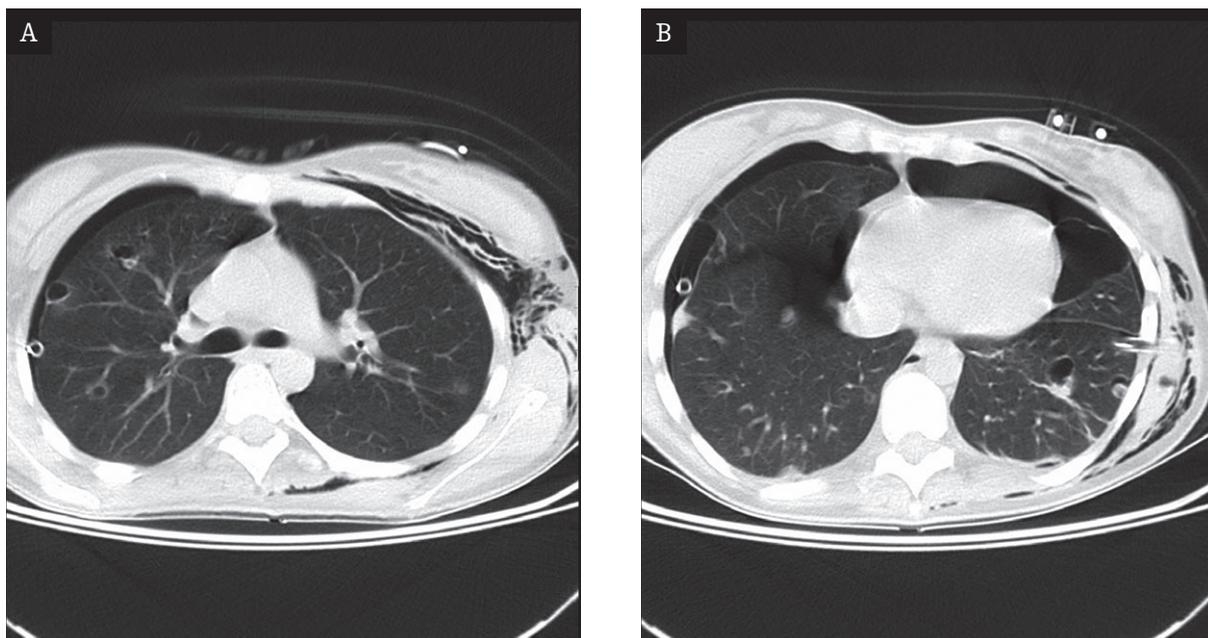


Figure 5 A, B. CT, image of the lungs after two months of chemotherapy.

interstitial, up to 1 cm in diameter. Apart from that, numerous bilateral solid foci or solid ones containing fluid, small interstitial infiltrates and atelectasis were also visible. The left lung was affected to a larger extent.

Taking into account the underlying disease, disintegration of the metastatic foci was primarily considered, to be differentiated from lesions of bacterial or mycotic etiology (fig. 4a and 4b).

Bronchoscopy revealed normal condition of the trachea and the bronchi, without purulent discharge, with single blood streaks. Fungal infection was also excluded.

After ca. 2 months of intensive drainage and chemotherapy, a slight reduction of the size and number of nodular lesions in the lungs was observed, as well as reduction of the pneumothorax (fig. 5a and 5b).

Discussion

The literature contains scarce descriptions of pneumothorax cases due to tumor metastases. In the aforementioned reports, the pneumothorax accompanied most frequently (although not exclusively [1]) tumors of sarcoma type:

- sarcoma synoviale of the lower extremity in a 33-year-old man – as the first symptom of pulmonary metastases [2],
- epithelioid sarcoma in a 39-year-old female patient with pulmonary lesions of thin-walled cyst type less than 1 cm in diameter and small nodules [3],
- angiosarcoma in a 69-year-old male patient with numerous thin-walled spaces in both lungs, which were resistant to treatment [4],
- leiomyosarcoma and sarcoma synoviale of the lower extremity in 2 patients with metastatic cystic pulmonary lesions, which caused pneumothorax [5],
- despite no evidence of metastatic lesions in a 11-year-old girl with osteosarcoma of the femur, they were suspected because of spontaneous onset of pneumothorax [6].

In the cited cases, like in our patient, small, solid pulmonary lesions were visible in classic plain X-ray. However, it is difficult to visualize cyst-like lesions, especially small and located peripherally, with this method, and their presence cannot be excluded. Thus, CT of the lungs is an examination allowing to assess the extent and stage of the lesions and

leading to the ultimate diagnosis. The treatment involves suction drainage improving the patient's respiratory efficiency, as well as complex causal therapy of the primary disease.

Traweek et al. [5] recommend in their report surgical removal of the metastatic foci which cause pneumothorax. If there are few metastases, and they are located in one lung, such management may yield a good effect. However, in case of multiple foci located bilaterally, thoracotomy and attempts of resection seem to be risky because of the burden of the procedure itself, as well as probability of low effectiveness of the surgery. A question arises, whether or not will the foci visualized on CT as solid cause a recurrence of pneumothorax after some time.

The presented case as well as those described in the literature suggest that in the situation of recurrent pneumothorax, treated so far as spontaneous (most often in young patients), CT of the lungs is recommended to exclude organic causes, especially pulmonary metastases presenting as the first symptom of neoplastic disease.

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