

Lung metastasis from tibialadamantinoma

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ABSTRACT

Lung metastasis from tibialadamantinoma

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Adamantinomas are rare primary low grade malignant bone tumors' (<1% of all bone cancers), which are usually located in tibial diaphysis. We present a case of lung metastasis in a 45 year old female who underwent left knee amputation because of tibial adamantinoma.

INTRODUCTION

Adamantinomas are rare primary low grade malignant bone tumors' (<1% of all bone cancers), which are usually located in tibial diaphysis.

CASE PRESENTATION

A 45 year-old female, non smoker, presented at our pneumonology department with symptoms of thoracic pain and cough without fever. She complained for dyspnea and fatigue on minimal exertion for the last 2 months. Physical examination and laboratory results did not reveal anything abnormal. Chest radiography demonstrated a large mass within the left lung perihilar

region (Figure 1). She had a history of left over-knee amputation because of tibialadamantinoma diagnosed 6 years before. Because of suspected lung metastasis additional imaging studies were performed: Brain, Abdomen CT scans and Chest CT scan which confirmed the left well circumscribed rounded perihilar tumor and moreover revealed secondary metastatic lesions in left lower lobe in contact with pleural surface. Osteolytic lesions located in left 3rd-5th ribs were demonstrated by both Chest CT scan and bone scintigraphy exam. No local bone recurrence was detected.

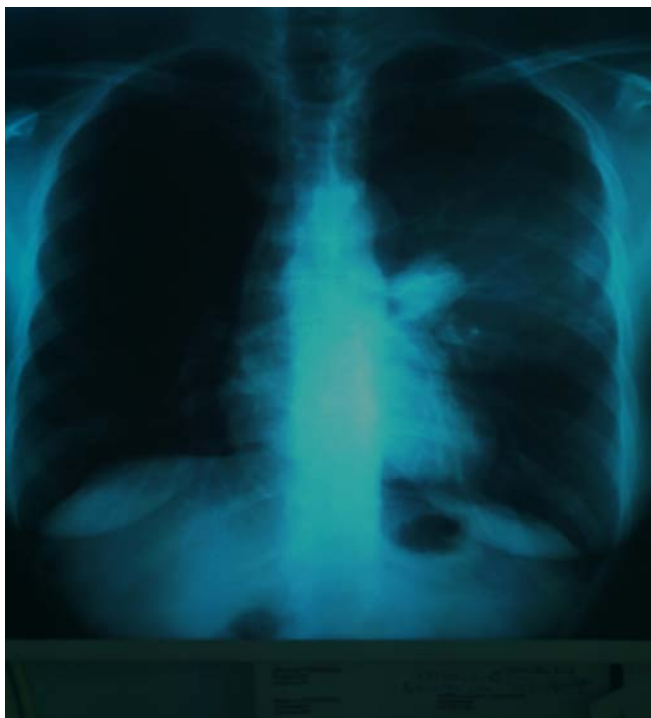
FibreopticBrochoscopy was performed and samples from transbronchial biopsies from the central mass sent to the lab for diagnosing testing.

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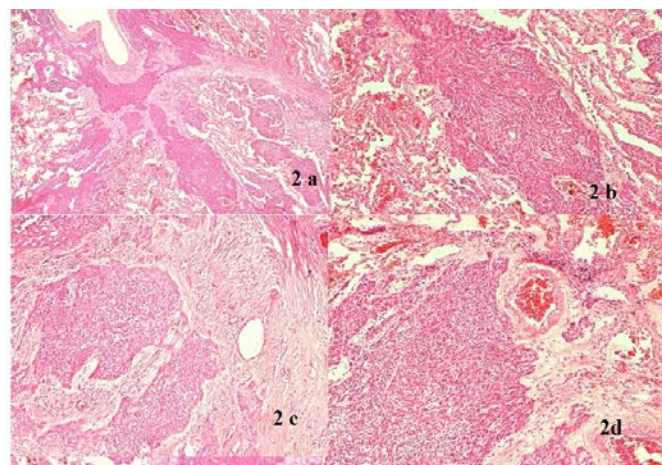
Microscopic findings were suggestive for lung metastatic adamantinoma. More specifically, the histological examination showed infiltration of the lung parenchyma by a malignant neoplasm, consisting of basaloid cells in a connective tissue stroma. The neoplastic cells were medium-sized, polygonal to spindle-shaped, with eosinophilic cytoplasm and hyperchromatic nuclei, lacking significant nuclear pleomorphism. The cells formed sheets, cords and whorl-like structures, quite often surrounding blood vessels and nerves, with characteristic palisading of the peripheral cells and evident reverse polarity of the nuclei away from the basement membrane. The neoplasm involved sites of necrosis and hemorrhagic foci (Fig. 2).

Figure 1. Preoperative x-ray revealing the left perihilar tumor



Based on the histopathologic results an extended lung resection under general anesthesia was decided. As the procedure was elective, two hours before, the patient received 5 mg diazepam. In the operating theatre, under hemodynamic monitoring (electrocardiogram, SpO₂, invasive arterial and central venous pressure monitoring) the anesthesia was induced with 150 mg propofol, 0,5 mg fentanyl and 70 mg rocuronium.

Figure 2. Histopathologic findings



2a. Anastomosing sheets and cords of neoplastic cells within desmoplastic stroma, infiltrating lung parenchyma. (H-E stain x40) **2b.** Polygonal to spindle-shaped basaloid epithelial neoplastic cells with eosinophilic cytoplasm, forming a whorl-like structure. (H-E stain x100) **2c.** Two aggregates of tumor cells, with characteristic palisading of the cells at the periphery, surrounded by desmoplastic connective tissue stroma. (H-E stain x100) **2d.** A sheet of tumor cells, surrounding a blood vessel. Markedly hemorrhagic foci. (H-E stain x100)

The patient was intubated with right double endotracheal tube No 39, in order to ventilate the right lung. During the procedure, the patient

received IV propofol and remifentanyl. She was ventilated with FiO_2 1 and TV of 6-8 ml/Kg with 14-18 breaths/min to achieve normocapnia.

The procedure lasted about two and a half hours and patient was infused with about two liters R/L. Her urine output was satisfactory. She underwent left pneumonectomy en block with a part of invaded pericardium with concomitant chest wall resection: posterior part of 3rd to 5th ribs were resected.

She was extubated two hours later in the ICU. Her postoperative course was uneventful. Patient died 2 years after the procedure suffering from multiple spinal bones metastases.

DISCUSSION

Adamantinoma of long bones or extragnathic adamantinoma represent a quite rare histologic type of slow-growing tumor. This type of tumor represents 0.1-0.5% of all primary bone neoplasms and characterized by low incidence of metastasis. The tumor is of epithelial origin. Differential diagnosis from other histologic type is not always easy¹. It should be distinguished from chondromyxoid fibroma, osteofibrous dysplasia, fibrous dysplasia and chondrosarcoma. Adamantinoma was first described by Fischer on 1913². Adamantinoma of bones usually affects tibia in 80% of the cases in diaphysis area in particular but it may also affects fibula, femur and ulna. There 4 different histo-

logic types: basaloid, fusiform, malpighian and tubular³. Another classification for adamantinomas includes the following two types: classic and differentiated type. In classic type patients are older than 20 years old while histologically the tumor involves epithelial and osteofibrous tissue. In differentiated type, patients are usually younger than 20 years old and osteofibrous tissue is the main component. Osteofibrous dysplasia- (OFD)- like adamantinoma represents a form of the differentiated type. In clinical cases swelling and pain are the main symptoms. Pathologic fractures have also been reported. Bone adamantinomas has many histological similarities with mandible adamantinoma or ameloblastoma, a tumor of odontogenic epithelium. These neoplasms are more common in men. Only 10-15% of adamantinomas are complicated with metastatic disease⁴. Distant metastasis develops many years after the first presentation. The usual metastatic sites are lungs, lymph nodes, liver, other bones and pericardium while local recurrence is not unusual. Radiographic imaging characteristics of this tumor are the following: well-demarcated, usually eccentric, with lobulated osteolytic lesions giving a characteristic 'soap-like' appearance, without any periosteal reaction. Local extension to the soft tissue may be seen. Its treatment should be exclusively surgical with wide en-block resections or amputation of the affected bone. MRI should be performed before

resection in order to define the surgical resection borders. Adamantinoma is locally aggressive and the local recurrence rate is very high. Lung metastases may be presented as spontaneous pneumothorax because of cystic lesion development as that has been described in 2 cases by Gonçalves et al.⁵. Perez –Ordonez and Bedard described the histological features of 2 cases of lung metastatic adamantinomas. Electron microscopy and immunocytochemistry with positive staining for keratin and vimentin are essentials for the diagnosis as well as a history of bone adamantinoma⁶.

Primary adamantinoma is resistant to radiation therapy and chemotherapy. In metastatic lung adamantinomas positive response in chemotherapy has been reported in few cases. Cohen et al found that therapy with pazopanib showed good results in lung metastatic adamantinoma⁷. Similar results have also been reported by Liman et al, after therapy with sunitinib⁸. Both pazopanib and sunitinib are multi-tyrosine kinase inhibitors.

CONCLUSION

Although the incidence of tibial adamantinoma metastasis is rare, its late development cannot be excluded. Histopathologic features of biopsy samples in combination with a history of bone adamantinoma are essential in documenting lung metastasis.

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Keywords: Lung tumor, Tibialadamantinoma, Lung metastasis

Author Disclosures:

Authors Ampatzidou F, Pasteli N, Voulioti E, Madesis A, Kilmpasani M, AsteriTh have no conflicts of interest or financial ties to disclose.

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