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Ewing's Sarcoma of the Foot with Multiple Synchronous Lung Metastases

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Abstract

***Introduction:** The foot isexceptionally concerned by Ewing's sarcoma (ES) and only a few sporadic cases of tarsal bones involvement have been reported in the literature.*

We are reporting an original observation of tarsal ES with significant locoregional spread to other bones and soft tissues of the foot and multiple synchronous lung metastases.

***Case report:** The 13-year-old boy, with no pathological medical history, was admitted for left foot pain evolving for two months, and with progressive aggravation leading to functional left lower limb impotence.The somatic examination noted a painful left foot on palpation and the slightest mobilization, with a hard and sensitive mass at the level of the sole of the foot.Biology showed a marked inflammatory syndrome. Radiological investigations and local biopsy concluded to the diagnosis of foot ES involving tarsal and metatarsal bones with multiple and bilateral synchronous pulmonary metastases. The child was referred to pediatric oncology department for chemotherapy.*

***Conclusion:** ES remains a rare primitive malignantbonetumor. The location at the foot is rare and tarsal involvement is exceptional. Pulmonary metastasis present at the moment of the diagnosis of ES is a major prognostic factor.*

Keywords: *Ewing's sarcoma,tarsal bones,bone sarcoma, foot, lung metastases.*

Introduction

First described in 1921 by James Ewing[1], Ewing's sarcoma (ES) is a primary malignant bone tumor of neuroectodermal origin that is particularly common in children and adolescents, and is characterized by its high grademalignancy, osteolytic character, and locoregional aggressiveness[1,2].

It can be seen in all the bones, but the preferred seats remain the long bones (60% of the cases)[2,3]. The short bones, especially those of the foot remain exceptionally concerned by this neoplasia[3,4].

Only a few sporadic cases of ES of tarsal bones have been reported in the literature[5-9].

We are reportingan original observation of tarsal ES with significant locoregional spread to other bones and soft tissues of the foot and multiple synchronous lung metastases.

Case Report

The 13-year-old boy, with no pathological medical history, was admitted for left foot pain evolving for two months, and with progressive aggravation leading to functional left lower limb impotence.The patient did not report local trauma or anterior skin lesion of the foot or ankle.

The patient's general condition was impaired with marked asthenia, significant recent weight loss, and fever at 38°C.

The somatic examination noted a painful left foot on palpation and the slightest mobilization, with a hard and sensitive mass at the level of the sole of the foot.

Biology showed a marked inflammatory syndrome withan erythrocyte sedimentation rate at 65mmH1, a C reactive protein at 22mg/l and a polyclonal

hypergammaglobulinemia at 28g/l, and without any other abnormalities.

The standard radiograph of the forefoot noted extensive osteolysis of the tarsal bones. The computed tomography of the foot concluded to an extensive, heterogeneous, and osteolyticlesion of tarsal and metatarsal bones of 6×3 cm of diameterwith intra-tumor calcifications (**Fig 1**). There was noted a marked invasion of adjacent soft tissues and an important densification of subcutaneous fat (**Fig 2**).

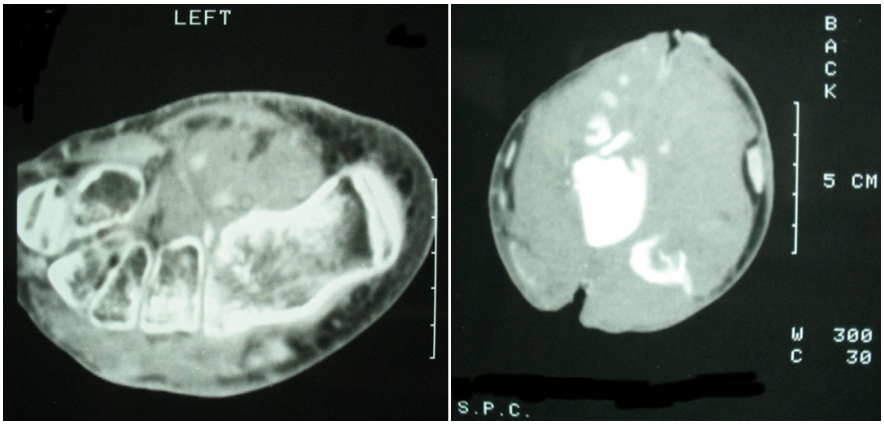

Fig 1. *Axial CT of the left foot: heterogeneous osteolytictumor of tarsus and metatarsal with calcification and invasion of soft tissues.*

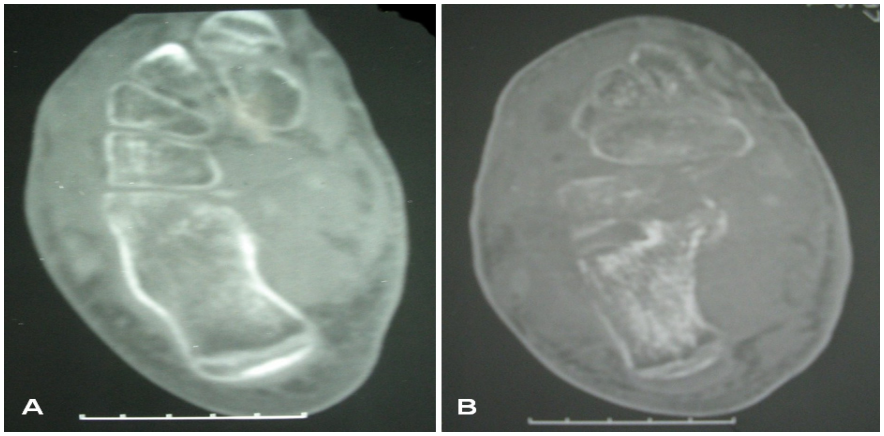

Fig 2. *Axial CT of the left foot in soft-tissue (A) and bone (B) windows: the importance of soft tissue invasion and osteolysis.*

The thoracic CT-scan revealed the presence of multiple and bilateral intra-parenchymal metastatic lung nodules affecting the posterior segment of the left upper lobe (**Fig 3**) and the apical segment of the right lower lobe(**Fig 4**).

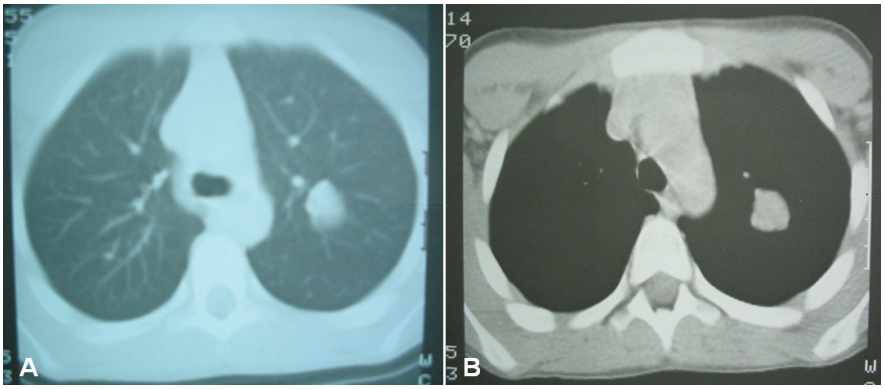

Fig 3. *Axial chest CT in parenchymal (A) and mediastinal (B) windows: metastatic pulmonary nodule of the posterior segment of the left upper lobe.*

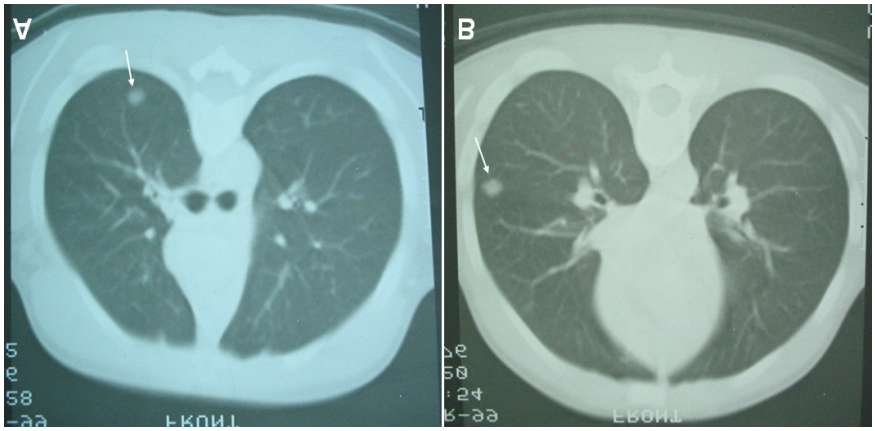

Fig 4. *Axial chest CT in parenchymal window: multiple metastatic pulmonary nodules of the apical segmentof the right lower lobe.*

Local biopsy was performed and the histopathological exam was compatible with ES. The diagnosis of Ewing's sarcoma of the foot with multiple synchronous pulmonary metastases was retained and the child was referred to pediatric oncology department for chemotherapy.

Discussion

Malignant bone tumors generally remain exceptional and unusual in the foot and ankle: only 54 cases in the large series of 8,542 bone tumors of Dahlin and Unni, corresponding to a frequency of 0.6% [10].

ES is also a very rare malignant tumor; its incidence in the general population is estimated at 1 case / 2-3 million / year [10]. It is particularly common in children and adolescents in whom it is the second primary malignant bone tumor, accounting for 5 to 15% of all these tumors [9].

Foot involvement during ES is rarely reported [3,10]. Indeed only 16 patients were recruited over a 38-year period in the Adkins CD et al series [4]. The overall frequency of this location is estimated at only 5% of all ES [11].

These localizations are characterized by the difficulties and the important delays to make the initial diagnosis [3] and by a treatment not yet consensual with a lot of controversies [10].

Tarsal bones are rarely affected by this sarcoma and only a few sporadic cases have been reported in the literature [5-10].

The clinic of this location is not specific compared to other locations of ES. It is often summarized as localized pain, bone swelling and/or a palpable mass of adjacent soft tissues [5-9]. Similarly, the radiological features usually observed in classical ES of long bones are also noted in the bones of the tarsus, particularly; bone lyticdestruction, cortical violation, aggressive periosteal reaction, and soft tissue mass [6].

However, it has been reported that ES of tarsal bonesis more often characterized by atypical radiological findings that explain the delayed diagnosis of these localizations [6].

The frequency of pulmonary metastases at the time of diagnosis of ES of the foot is variously estimated according to the series: it varies from 1 patient / 14 (7.14%) in the series of San-Julian M et al [10] to 7 patients / 16 (43.75%) in the series of Adkins CD et al [4].

The presence of these metastases at the time of diagnosis represents a pejorative element for the prognosis of ES. Indeed, only one of the seven patients with ES and synchronous pulmonary metastasis in Adkins CD et al series survived in contrast to eight out of nine patients with localized ES without metastases [4]. Similarly, in Casadei R et al series, 100% of patients with ES and metastases at the time of diagnosis had died, contrasting with 44% survival at an average seven-year follow-up interval among those with localized form of ES without pulmonary metastasis at the time of diagnosis [12].

The treatment classically combines radiotherapy and surgery for local control of neoplasia, and chemotherapy for its systemic control, particularly if metastases [4,10]. Chemotherapy has revolutionized the management of ESand significantly improved its prognosis [3,10]. Amputations can be indicated for unresectable tumors [10].

Conclusion

ES remains a rare primitive malignant bone tumor. The location at the foot is rare and tarsal involvement is exceptional. Pulmonary metastasis present at the moment of the diagnosis of ES isa major prognostic factor. Our observation is distinguished by the localization in the tarsalbones, and its association with multiple and bilateral synchronous pulmonary metastases.

Early diagnosis and adequate management are the only guarantors of a better prognosis for Ewing's sarcoma.

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