CASE REPORTS

PLEURAL METASTASIS OF A PEDIATRIC OSTEOSARCOMA

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Abstract

Introduction: *Osteosarcoma is the most common primary bone tumor in children and young adults.*

Although osteosarcoma is a tumor with a great metastatic potential, mainly to the lung; pleural metastasis in patients with osteosarcoma are rarely reported.

We present a case of 16 years-old male with a pleural metastasis of a tibial osteosarcoma diagnosed 4years earlier. He was submitted to a left thoracotomy and intra-operatively a pleural mass and a left upper lobe lesion was identified. Video-assisted resection of the extra-pulmonary mass and a wedge resection of the left upper lobe lesion was performed. The surgery was uneventful.

The patient is clinically well, asymptomatic, maintains active surveillance.

Keywords: *Osteosarcoma, pleural metastasis, pediatrics*

INTRODUCTION

Osteosarcoma is a form of primary bone tumor thought to derive from primitive mesenchymal bone-forming cells. It is the most common primary bone tumor in children and young adults, but it can also occur in younger children and older adults[1].

At presentation, 80-90% of the patients have localized disease and 10-20% have metastasis[1,2]. Several prognostic factors affecting overall survival have been identified in patients with osteosarcoma, such as tumor location and size, metastasis, surgical resectability, and degree of tumor necrosis after neoadjuvant chemotherapy[3,4]. Osteosarcoma is characterized by its strong metastatic potential; approximately 25-30% of the tumors recur after treatment and the lung and other bones are the most common sites of initial recurrence. Pleural metastasis in patients with osteosarcoma is rarely reported. Pleural metastasis can occur due to direct contact of pleural with the lungs, or by hematogenous spread[1-3].

CASE REPORT

We present a case of a 16 years-old male patient with a right-side tibial osteosarcoma diagnosed in March of 2016. He underwent in accordance with the EURAMOS (European and American Osteosarcomas Studies) protocol, with neo-adjuvant chemotherapy, surgery with extended resection and reconstruction with heterologous structural tibial graft and tibiocalcaneal arthrodesis, and adjuvant chemotherapy. Histopathological examination of the lesion showed a viable area of less than 10%, and

soft tissue and bone margins free of neoplasia. There were no major complications and remission at the end of the treatment was confirmed.

In September of 2020, the patient presented with an asymptomatic hypotransparency in left hemithorax. Contrast-enhanced computed tomography of the thorax revealed an extrapulmonary mass of pleural/chest wall origin, next to the upper lobe of the left lung. This was described as a heterogeneous lesion, with 93x51x84mm, most likely corresponding to a secondary lesion (Figure 1). There was no identifiable lesion in the lung parenchyma and no pleural effusion. Positron Emission Tomography (PET) revealed a hypermetabolism of 18F-FDG in this large lesion, next to the upper third of the left pulmonary field, adjacent to the chest wall. The aspiration biopsy of the lesion revealed fragments of malignant, pleomorphic, high-grade mesenchymal neoplasia. These aspects were suggestive of osteosarcoma metastasis. Magnetic resonance imaging of the right leg was also performed, with no evidence of expansive masses or suspicious contrast uptake.

A left posterolateral thoracotomy was performed, and a pediculate mass of approximately 11 cm was identified on the left parietal pleura, unrelated to the lung (Figure 2). A second pleural mass next to the lesion (Figure 3) and a millimetric nodule in the left upper lobe was also identified (Figure 4), as well as a small pleural effusion.

Video-assisted resection of the extra-pulmonary mass was performed, using ultracision®, as well as of the pleural nodular lesion. A wedge resection of the left upper lobe including the nodule was performed with GIAS® 55mm. After careful inspection and palpation, no other nodules were detected. Histopathological examination from all the lesions was suggestive of metastatic osteosarcoma. Tumor giant cells with large hyperchromatic nuclei were present, showing osteoid formation. Extensive areas of the tumor exhibited chondroid differentiation along with mitotic figures. Cytology of the pleural fluid revealed the presence of reactive mesothelial cells and some inflammatory cells. The surgery was uneventful.

Post-operative CT (14 days after surgery) revealed a nodular heterogeneous density area with 56x33x26mm compatible with a serohematic collection only.

After one year of follow up, the patient is clinically well and asymptomatic. He maintains follow-up with us, with clinical and imaging surveillance.

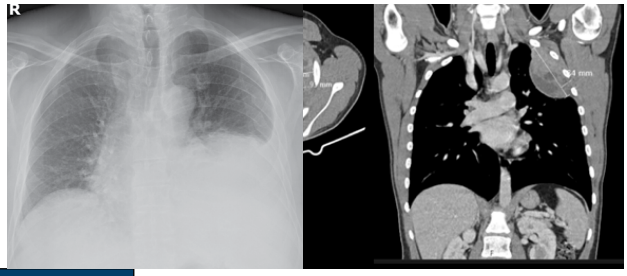

Figure 1 *Contrast-enhanced computed tomography of the thorax revealed an extra-pulmonary mass of pleural/chest wall origin, next to the upper lobe of the left lung, with 93x51x84mm.*

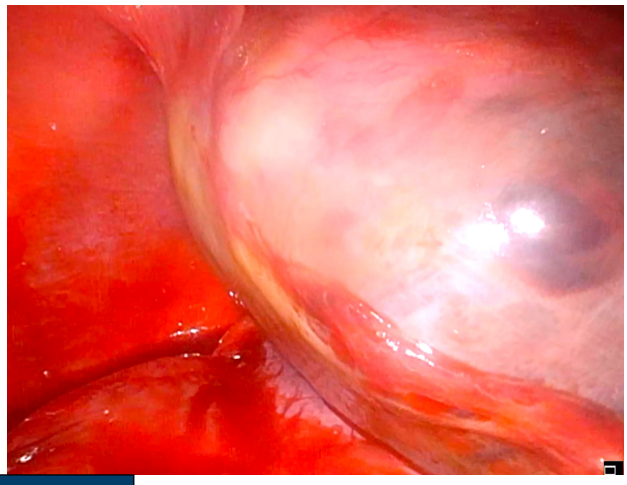

Figure 2 *Pediculate mass of approximately 11 cm identified on the left parietal pleura, unrelated to the lung.*

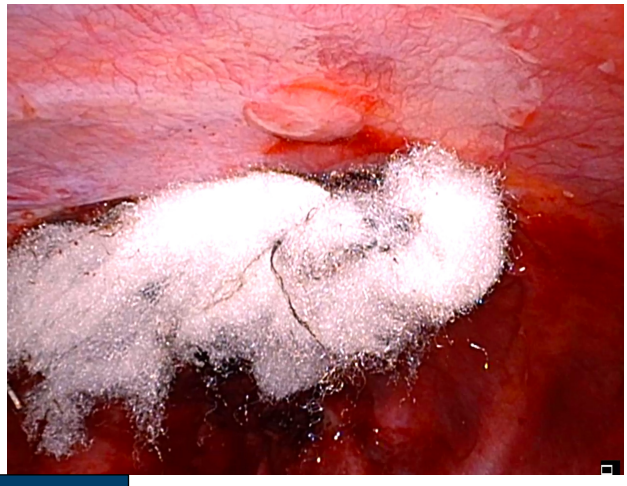

Figure 3 *Pleural mass, next to the main lesion.*

DISCUSSION

Osteosarcoma is a deadly form of primary bone tumor thought to arise from primitive mesenchymal bone-forming cells. It is the most common primary malignant neoplasm of the bone with over 60% of the cases occurring in patients 10-20 years old[5]. At presentation, 80% of the patients have localized disease and 10% have distant metastasis[6]. Recurrent osteosarcoma in general

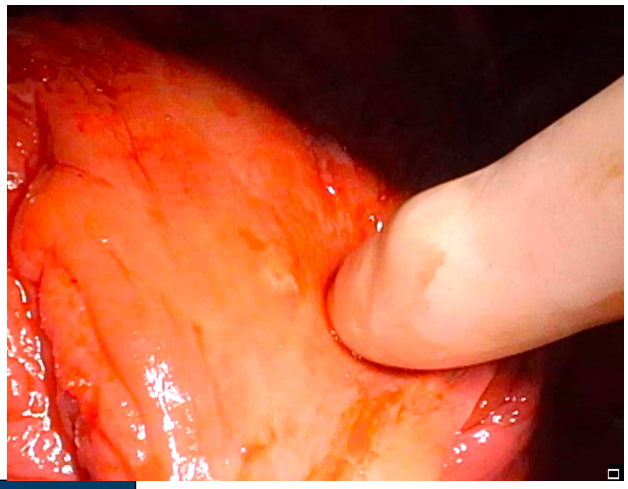

Figure 4 *Nodule in the left upper lobe.*

terms is considered to have a poor prognosis. A short time-interval between initial presentation and relapse, recurrences involving more than one site, and in the case of lung metastases, bilateral disease, and pleural involvement are negative prognostic factors[1]. The most common sites of metastases are lung and bone, respectively. According to the Japan Autopsy Annual Database, 643 patients died of osteosarcoma between 1981 and 2002 in Japan, of whom, only 78 (12.1%) patients had pleural metastases[7].

Overall management and prognosis in cases of metastatic osteosarcoma is determined by the number, site and size of the metastases. In recurrent osteosarcoma presenting with lung metastasis or pleural effusion; the pleural cavity must be thoroughly examined with a thoracoscope. After direct visualization of the pleural cavity, if any metastatic parietal pleural lesion is found in contact with the lung lesion or isolated, it should be resected completely along with the lung tumor and free pleural margin. Complete surgery is a prerequisite for cure and improves the survival of the patient[8-10].

REFERENCES

1. Saha D, Saha K, Banerjee A, Jash D. Osteosarcoma relapse as pleural metastasis. South Asian J Cancer. 2013;2(2): 56. doi: http://doi.org/10.4103/2278-330X.110483:10.4103/2278-330X.110483
2. Li W, Zhang S. Survival of patients with primary osteosarcoma and lung metastases. J Buon. 2018; 23(5): 1500-1504
3. Marchiori E, Barreto M, Zanetti G. Pleural Metastasis of Osteosarcoma. Images in Cardiothoracic surgery. 2018; 105: E87-E88. doi: https://doi.org/10.1016/j.athoracsur.2017.09.010
4. Sujing J, Guannan W, Ying D. Endostar combined with chemotherapy in a pediat-ric osteosarcoma with pulmonary metastasis and malignant pleural effusion: A case report. Medicine. 2017;96(51): 9077. Doi http://dx.doi.org/10.1097/MD.0000000000009077
5. Krishnamurthy K, Alghamdi S, Kochiyil J, Bruney G, Poppiti R. Osteosarcoima pre-senting with malignant pleural effusion in a 55 year old, Respir Med Case Rep. 2018;25:314-317. Doi 10.1016/j.rmcr.2018.10.017
6. Marina NM, Pratt CB, Rao BN, Shema SJ, Meyer WH. Improved prognosis of children with osteosarcoma metastatic to the lung (s) at the time of diagno-sis. Cancer. 1992;70:2722–7.
7. Japan Autopsy Annual Database. Japanese Society of Pathology; Tokyo: 1981-2002. (Japanese Society of Pathology. In Japanese).
8. Salah S, Toubasi S. Factors predicting survival following complete surgical remission of pulmonary metastasis in osteosarcoma. Mol Clin Onco. 2014; 157-162. doi: https://doi.org/10.3892/mco.2014.426
9. AbdullGaffar B. Osteosarcoma pleural effusion: A diagnostic challenge with some cytologic hints. Diagn Citopathol. 2021;49(1): E40-E41. doi: https://doi.org/10.1002/dc.24569
10. kim S, Choi J, Lee S, Choi J, Hong S, Chung H, Kang H. Imaging findings of extrapulmonary metastases of osteosarcoma. Clin Imaging. 2004; 28(4): 291-300. doi: https://doi.org/10.1016/S0899-7071(03)00206-7.