Chest Wall Metastasis Resulting from a Primary Extremity Osteosarcoma- a Dramatic Clinical Presentation

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Sir,

Osteosarcoma is the most common primary malignancy of the bone, with a peak incidence in the second decade and an additional peak in the elderly individuals (seventh and eighth decades) [1]. Improvements in multimodal treatment for osteosarcoma, especially in the use of neoadjuvant and adjuvant chemotherapy have increased the disease-free and the overall survivals. Meanwhile, the prolonged survival of patients has permitted the appearance of new, significant, extra pulmonary targets for metastasis.

A 12-year-old boy presented to us with an expansible swelling over the left distal femur, a Jamshidi needle biopsy was suggestive of a high grade osteosarcoma. His initial metastatic work up (CT chest and bone scan) was normal. He received six cycles of ifosfamide, doxorubicin and cisplatin based chemotherapy (4 neoadjuvant and 2 cycles of adjuvant) and a limb salvage surgery with a custom based megaprosthesis. The histopathology of the resected tumor showed residual osteosarcoma with 90 % necrosis. Four months following completion of his adjuvant chemotherapy, he presented to us with a rapidly increasing left anterior chest wall swelling over 2 weeks duration (Fig. 1a). A CT chest revealed an irregular 4×2 cm mildly enhancing mass lesion eroding the left 4th rib, the underlying pulmonary parenchyma was however normal (Fig. 1b). His loco-regional disease was clinically under control as was also

confirmed by a bone scan. A trucut biopsy from the chest wall swelling was suggestive of metastasis from osteosarcoma (Fig. 2a, b). In view of the short disease free interval and a rapid progression of the chest wall swelling; he was planned for upfront chemotherapy using high dose methotrexate, with a plan to incorporate a surgical resection based on the response. The chest wall lesion unfortunately progressed while the patient was on the second cycle of chemotherapy to a size of 9.3×8.4×7.7 cm. It was found to infiltrate the pericardium and the thymus; in addition multiple bilateral pleural and sub pleural based nodules were seen in both the lung field, suggestive of lung metastasis as seen in the PET-CT (Fig. 3c, d). The patient refused to accept any additional intensive chemotherapy and was discharged on oral chemotherapy with single agent etoposide.

One-third of patients with osteosarcoma will present with recurrent/metastatic disease during the course of their disease [2]. The lung is the most common site of metastases, with 77 % to 92 % of patients experiencing recurrence at this site and it is reported to be the only site of metastasis in nearly two thirds of the patients. The most common site of the extra pulmonary metastases is bone; the unusual sites of metastasis include the brain, abdominal viscera, lymph nodes, pleura, pericardium and oral cavity. Surgery is an essential component of therapy in the relapse setting; multi-agent chemotherapy may contribute to the improvements in the survival outcomes. A more aggressive clinical behavior seems to be a feature in these patients in whom the prognosis is extremely poor [3, 4], the average survival after recurrence is generally less than a year. In conclusion, considering the dramatic clinical presentation of our patient, extra pulmonary metastasis should be considered as a differential in patient's previously diagnosed with a primary osteosarcoma.

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Fig. 1 **a** Clinical presentation of the rapidly increasing (4×2 cm) left anterior chest wall swelling four month following adjuvant chemotherapy. **b** A CT chest showing an irregular 4×2 cm mildly enhancing mass lesion eroding the left 4th rib

Fig. 2 **a, b** H & Ex20- Section shows tumor fragments composed of sheets of round to oval cells with moderate cytoplasm and vesicular nuclei with prominent nucleoli, occasional tumor giant cells in an osteoid matrix, suggestive of metastatic osteosarcoma high grade

Fig. 3 **a, b** Clinical photograph demonstrating the progression of the left anterior chest wall swelling (9×8 cm) on salvage chemotherapy. **c, d** PET-CT showing progression of the chest wall swelling to attain a size of $9.3 \times 8.4 \times 7.7$ cm; in addition a sub pleural based nodule is seen

Competing Interests None declared

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Ethical Approval Not required

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