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Metastatic osteosarcoma of the ethmoid: an unusual cause of recurrent epistaxis and proptosis

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Abstract
Osteosarcoma is the most common primary malignant tumour of bone and it usually metastasises to the lung. In the nasal cavity metastatic disease is extremely rare. We describe a case of osteosarcoma presenting with recurrent epistaxis, and proptosis due to secondaries in the nasal cavity. To our knowledge such a case has not been reported previously in the available English literature.

Key words: Osteosarcoma; Neoplasm metastasis; Nasal cavity; Epistaxis; Exophthalmos

Introduction

Metastatic disease to the nose is very rare with fewer than a hundred instances having been recorded in the literature (Johnson *et al.*, 1993). Osteosarcoma is the most common primary malignant tumour of the bone and more than 90 per cent of patients with this disease die with pulmonary metastases (Jeffree *et al.*, 1975). We present a case of osteosarcoma with metastases to the ethmoid bone causing recurrent epistaxis and proptosis.

Case report

A 15-year-old Malay male was referred from a district hospital for a swelling over the left thigh of three months duration. Open biopsy confirmed the diagnosis of osteosarcoma. He underwent disarticulation of the left hip. Post-operatively he was given high dose methotrexate with leucovarin calcium as a rescue treatment. Seven months later radiological examination of the chest showed secondaries in the left lung for which he underwent lobectomy. Two more cycles of chemotherapy were given post-operatively which included bleomycin, actinomycin

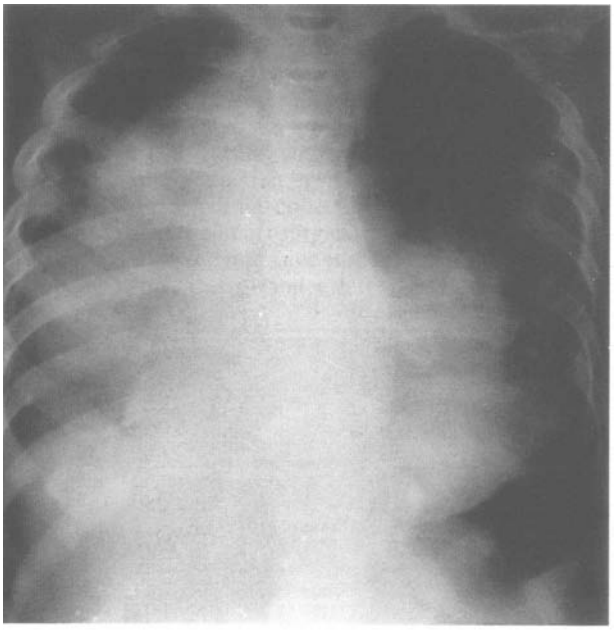

Fig. 1
Chest radiograph showing secondary deposits in both lungs.

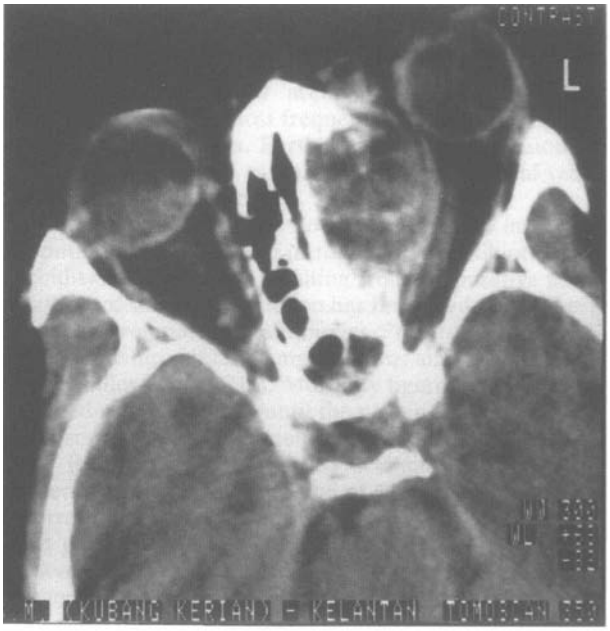

Fig. 2
CT Scan displaying metastatic deposit in the left ethmoid with proptosis of left eye.

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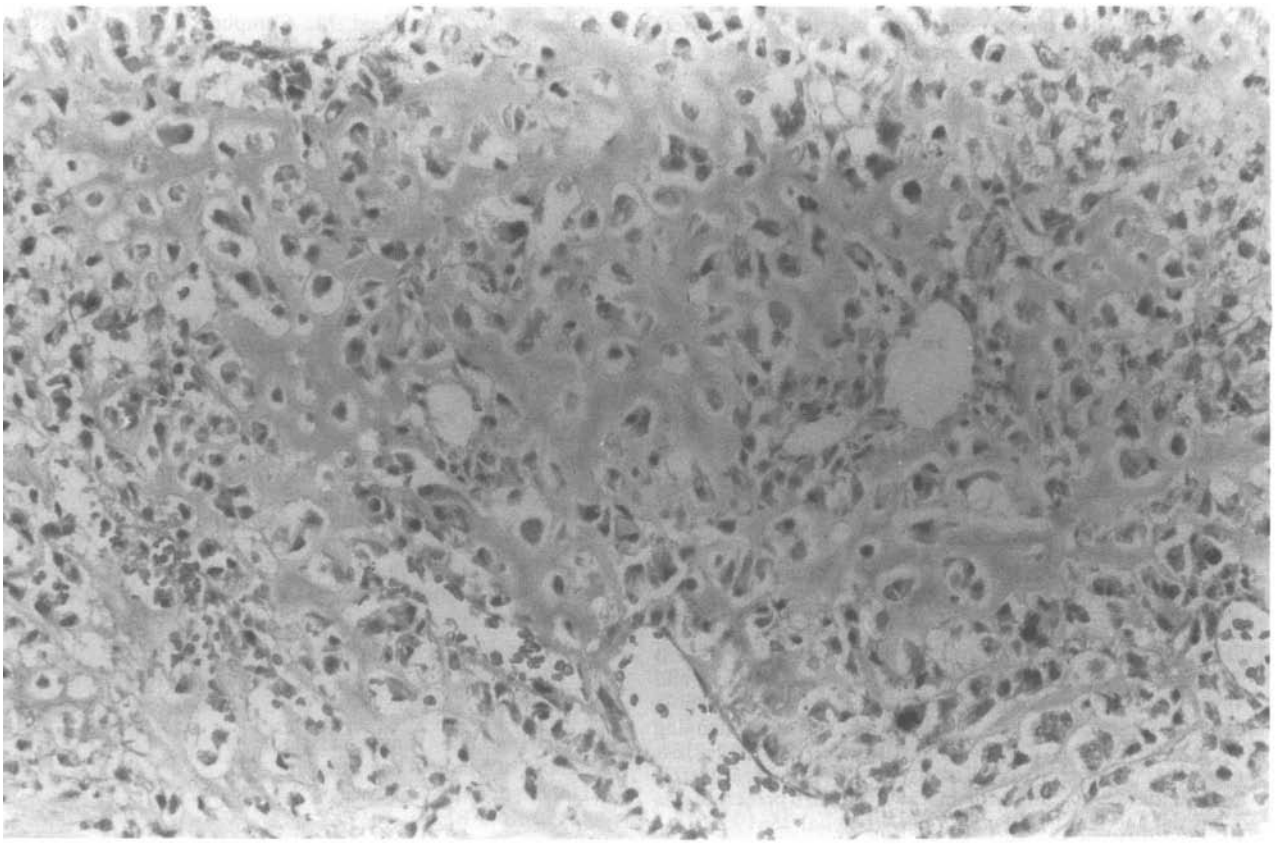

FIG. 3

Panoramic view of section from the nasal biopsy showing sheets of malignant cells with evidence of direct osteoid formation exhibiting 'Lace pattern' (H & E ×400).

and cyclophosphamide. Three months after the left lobectomy the patient developed secondary deposits in both his lungs (Figure 1). He refused further surgery.

Seven months later he was referred to the ENT Clinic for epistaxis. On examination he showed bleeding from the left nostril. The left nasal cavity was full of fresh blood clots and the left eye showed proptosis. He required repeated anterior nasal packings for control of epistaxis. Full blood examination revealed haemoglobin 8.5 gram/dl, with normal white cell and platelet counts. His computed tomography (CT) scan of the paranasal sinus showed a metastatic deposit in the left ethmoid (Figure 2).

Examination of the left nasal cavity under general anaesthesia showed a fleshy, vascular lesion in the lateral wall of the nose. Biopsy from this lesion showed the features of metastatic osteosarcoma (Figure 3). The patient's general condition deteriorated and he died after one month.

Discussion

Osteosarcoma (also known as osteogenic sarcoma) is the most common primary malignant tumour of bone. Histopathologically it is a primary malignant tumour of osteoblasts and is identified microscopically by direct formation of osteoid or bone, or both, by neoplastic cells. Most cases appear in children, adolescents and young adults with a male predominance. The classic site of occurrence is the medulla of the metaphysis of the long bones, particularly the lower end of the femur, upper end of the tibia and upper end of the humerus. Osteosarcoma is a highly invasive neoplasm but cartilage shows remarkable capacity to resist the invasion locally (Kissane, 1990). Blood borne metastases are seen mostly in the lungs (Jeffree *et al.*, 1975).

Recent studies indicate, however, that the incidence of nonpulmonary metastases is increasing. Giuliano *et al.* (1984) reviewed the clinical course of 111 patients with osteosarcoma for recurrence and reported pulmonary metastases in 61 per cent of patients. Sites of extrapulmonary metastases in order of frequency were bone in 14, epidural in three, brain in two and soft tissue, liver, mediastinal, diaphragmatic involvement in one patient each. Other rare extrapulmonary sites of metastases in osteosarcoma are larynx, (Shimizu *et al.*, 1994); kidney, (Kajikawa *et al.*, 1993); heart, (Dalal *et al.*, 1978; James *et al.*, 1993); adrenal gland, (Potepan *et al.*, 1992), and eyelid (Newman and Diloreto, 1987).

Metastases to the nose and paranasal sinuses are extremely rare and can occur from anywhere in the body occurring usually from the kidney and pancreas (Maran and Lund, 1990).

Epistaxis is a primary complaint in approximately 50 per cent of patients with secondaries in the nose and the figure may be still higher in more vascular tumours (Johnson *et al.*, 1993).

Radiographs of paranasal sinuses are not routinely done in patients with epistaxis because tumours have rarely been found to be a cause of epistaxis (Singh, 1992). Padgham (1990) reviewed 107 patients with epistaxis and concluded that routine use of X-ray to exclude neoplasm is neither necessary nor effective.

Our patient presented with recurrent episodes of unilateral epistaxis. This, together with the presence of proptosis, gave rise to a suspicion of secondaries in the nose. Radiological investigation showed the presence of

tumour in the ethmoid region which was later confirmed by biopsy as a metastatic osteosarcoma.

Conclusion

Recurrent epistaxis in a known case of osteosarcoma should be taken seriously. Besides doing basic investigations like full blood examination and a coagulation profile, one must also take radiographs of the nose and paranasal sinuses keeping in mind the possibility of metastases to these structures.

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