CASE REPORT

A rare case of giant cell tumour (GCT) of bone with lung metastases

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SUMMARY

A case of 16-year-old girl with giant cell tumour of right fibula is presented to us with bilateral lung metastases. In view of widespread bilateral lung metastatic lesions, the patient was given multimodality treatment. Chemotherapy followed by radiotherapy to the local site as well as lung bath has been given and has shown good response.

BACKGROUND

Giant cell tumour (GCT) of bone is a primary intra-medullary tumour which is supposedly benign but can be locally aggressive and even metastatic.¹ Its name originated from the giant cells found within the tumour. These giant cells show positivity for certain specific markers, including carbonic anhydrase II, tartrate-resistant acid phosphatase, cathepsin K and receptor activator of nuclear factor- κ B.²

GCT of bone accounts for 21% of all benign skeletal tumours and 5% of primary bone tumours.³ It has the highest prevalence among the population aged between 20 and 40 years old and has female to male ratio of 1.3:1.0 with slight female preponderance.⁴ The common sites for GCT include distal femur, proximal tibia and distal radius. Metastasis in GCT of bone varies between 0% and 9%.⁵ The most common sites include lungs. Other less common sites include brain, kidneys, bone, skin and lymph nodes.⁵ The present study describes a

case of GCT of right fibula who developed lung metastases 8 months after undergoing surgery for the primary lesion.

Pulmonary metastases in GCT of bone are rare. This case is being reported due to a good response of chemotherapy and radiotherapy in case of metastatic GCT which is quite unusual.

CASE PRESENTATION

A 16-year-old girl presented to us in March 2014 with chief complaints of pain and swelling in right lower leg for the past 3 months. She developed breathlessness and dry cough for the past 1 month. She was operated in September 2013 for the swelling at same site and complete excision was done. Biopsy report at that time has showed the lesion as osteoclastoma (GCT). No relevant medical, social or family history. On examination in March 2014, a diffuse lump was palpable at the scar site on lateral aspect of right lower leg just below the knee. On chest auscultation, diminished vesicular sounds were present over the bilateral chest area.

INVESTIGATIONS

Chest X-ray (March 2014) demonstrated the presence of multiple opacities in bilateral lung field suggestive of bilateral lung metastasis. MRI of the right knee joint done in March 2014 showed soft tissue component in region of excised segment of fibula representing mitotic soft tissue involvement ([figure 1A](#)). Histopathological report from the same site confirmed GCT. The histopathological details are described in [figure 2](#).

DIFFERENTIAL DIAGNOSIS

Osteoclastoma, Ewing's sarcoma and osteosarcoma.

TREATMENT

The patient was treated with six cycles of chemotherapy (adriamycin, ifosfamide and dacarbazine) followed by radiotherapy to the local site. Radiotherapy was delivered by Cobalt-60 Theratron machine by anteroposterior–posteroanterior (AP–PA) fields using source to skin distance technique. Radiation fields included the gross disease, the scar site, margins of 5 cm excluding the adjacent joints and a medial strip. Total radiotherapy dose of 60 Gy/30 fractions/6 weeks/5 fractions per week/2 Gy per fraction was given and it got completed on 10 November 2014. Chest X-ray done in December

Figure 1 (A) MRI of the right leg represents growth at excised fibular area. (B) MRI of the right leg represents post-radiotherapy fibrosis.

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Figure 2 Photomicrograph showing uniform and regular distribution of stromal cells and giant cells.

2014 showed resolution of lung metastases. She again came with breathlessness in March 2015 for which chest X-ray was done and it was suggestive of bilateral lung metastases ([figure 3A](#)). MRI of right knee and leg done in March 2015 has shown post op fibrosis and no contrast enhancement ([figure 1B](#)). She was further given three more cycles of chemotherapy followed by radiotherapy to the bilateral lungs (lung bath 15 Gy/10 fractions/2 weeks/5 fractions per week/1.5 Gy per fraction). Radiotherapy has been given by Cobalt-60 machine with AP-PA fields. Superior border of the radiation field extends above the superior border of the lateral edge of the clavicle, inferior border extends to level of L1 and lateral borders till areola of nipple, shielding the humeral head. Thereafter, X-ray showed complete resolution ([figure 3B](#)).

OUTCOME AND FOLLOW-UP

The patient was asymptomatic for 7 consecutive months with good subjective and objective responses. Thereafter, she lost to follow-up from October 2015 onwards.

DISCUSSION

Lung metastases in GCT was first reported in 1926 by Flinch and Gleave. The histology of pulmonary metastatic lesion is usually similar to the primary bone lesion.⁶ Histological findings include multinucleated giant cells and mononucleated stromal cells. Typical radiological findings include lytic lesion with trabeculation, no periosteal reaction and eccentric localisation in the metaphysis of long bones.⁷ Campanacci *et al* have classified

GCT into grades I, II and III with higher grades having more chances of recurrence and metastases.⁸ As in the present study, the GCT was of grade II; hence, aggressive course of the disease was somewhat predictable.

In a retrospective analysis on metastatic GCTs, it was found that lung lesions had appeared after the primary lesion had been excised, with time interval varying between 17 and 89 months. Also, lung metastases in GCTs are considered to have good survival rate. Thus, generally intense radiotherapy and chemotherapy is not considered due to the slow progression and the better survival rate of the disease.⁹ On the contrary, in the present case, the disease has metastasised within 6 months of the surgery for the primary tumour.

Treatment of GCT of bone is still controversial. Surgical treatment option for primary GCT of bone lesions includes intralesional surgery and wide resection. Intralesional curettage has a significantly higher local recurrence and pulmonary metastasis rate than wide resection. Radiation is the mainstay of treatment in inoperable and recurrent cases. It is also advocated in some cases as adjuvant treatment.¹⁰ Dose of the radiation which is required for treatment is around 60 Gy. Effective chemotherapy is given using chemotherapeutic drugs like Ifosfamide, cisplatin and epipodophyllin toxin for a short period. In a retrospective analysis by Dunst *et al*, it was seen that lung irradiation is beneficial in terms of improved survival of patients with pulmonary metastasis of Ewing's sarcoma.¹¹ In a cohort study, three patients of GCT with metastases to lungs received different chemotherapy regimens, including cyclophosphamide, doxorubicin and cisplatin (CAP), mesna, doxorubicin, ifosfamide and dacarbazine (MAID) and ifosfamide and etoposide (IFO+VP-16). None of these patients had achieved complete response, but still two out of the three patients had long-term survival.⁹ In another case of GCT with primary site being metacarpal bone, with multiple lung lesions, complete response was seen with chemotherapy regimen adriamycin + cisplatin (ADM + DDP).¹² In the present study, considering the aggressive nature of the tumour, combination chemotherapy regimen with Ifosfamide, adriamycin and dacarbazine has been started. Even if complete remission of pulmonary metastatic disease has been achieved in some studies after chemotherapy, prophylactic whole lung irradiation is recommended in all patients.¹¹

Figure 3 (A) Chest X-ray showing bilateral lung metastases. (B) Chest X-ray showing resolution of disease in bilateral lungs after lung bath.

Learning points

- Pulmonary metastases in giant cell tumour of bone may show good tumour control when treated with chemotherapy and radiation therapy.
- Lung bath (radiation to bilateral lungs) also has shown good disease control.

Contributors DKS: conception of the work, analysis, interpretation of data, drafting and final revision. SG: design of the work, analysis and interpretation of data. NB and SK: drafting and final revision.

Competing interests None declared.

Patient consent Guardian consent obtained.

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