Denosumab therapy for giant cell granuloma in a paediatric patient: using quantification of Tc99m-MDP uptake on SPECT imaging to guide treatment.

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INTRODUCTION

Giant cell granulomas (GCG) are uncommon bony lesions that most commonly affect the maxilla and mandible; whilst generally benign they can be disfiguring to the face. Histologically, GCGs are thought to be derived from osteoclast-like multinucleated cells. Despite being benign, they are often associated with localised bone destruction and in some cases, pain. They are commonly treated with surgical excision, radiation, or with medications such as bisphosphonates.

CASE PRESENTATION

An 11-year-old female presented with an oral lesion. CT scan and biopsy confirmed a diagnosis of GCG of the maxilla. Initial treatment with weekly steroid injections resulted in a short period of stability before growth progression returned. MRI/SPECT imaging showed an expansive, lytic left maxillary lesion with increased Tc99m-MDP uptake, measuring 36mm x 41mm x 42mm. Zoledronic acid was commenced, after which a small reduction in lesion size was noted, although MRI/SPECT imaging after a year of treatment showed no change with increased sclerosis and increased Tc99m-MDP uptake.

Denosumab was commenced at 60mg every 4 weeks (increased to 120mg after 11 months) by subcutaneous injection with calcium and vitamin D supplementation. The patient has maintained normal renal function and bone chemistry whilst on denosumab, bone density is within the normal range for age and no signs of osteonecrosis of the jaw. After 14 months of denosumab, the patient had suppressed bone turnover markers, MRI scan showed lesion size was unchanged but with increased sclerosis. However, semi-quantitative analysis showed a reduction in the ratio of Tc99m-MDP uptake in the lesion, compared to normal bone, from 4.9 to 1.4.

The lesion has clinically reduced after 19 months of denosumab with only mild facial asymmetry. Denosumab weaning will be gradual to avoid rebound hypercalcaemia after treatment cessation. In line with case reports in the literature, the patient will then receive two doses of zoledronic acid. Ongoing 3-6 monthly SPECT scans will help to guide whether denosumab needs to be restarted in the future.

CONCLUSIONS

This case report adds further support for the use of denosumab in the treatment of GCG in paediatric patients. Imaging using SPECT with semi-quantitative analysis of Tc99m-MDP uptake allowed for valuable assessment of denosumab effect which was difficult to appreciate on standard MRI/SPECT images.

REFERENCES

5. Joanne Blair FM, Carvalho NPF, Santos Tobi Aderotimi. Explore other case reports in children and potential side effects.

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