

## CASE REPORT

# Multiple craniofacial osteomas: an isolated case

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### Keywords

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**Abstract** Osteoma is a benign bony outgrowth of membranous bones and is found mostly on skull and facial bones. In the head and neck region they are commonly seen in the fronto-ethmoidal regions. The lesions are usually asymptomatic and sometimes may impinge on surrounding areas and cause symptoms. Osteomas may be present in a solitary or multiple sites. The present article reports a case of multiple osteomas of head located in the craniofacial region with imaging details.

## Introduction

Osteomas are benign, bone-forming tumours located within bones or developing on them (Meher *et al.*, 2004). They usually cause cosmetic deformity but depending on the location can also cause symptoms like headache, localised pain, hearing loss and vertigo (Marlowe *et al.*, 1980). The osteomas can be central, peripheral or of an extraskeletal type (Woldenberg *et al.*, 2005). Osteomas have a predilection for the head and neck region which includes the facial bones, skull, and mandible and is the most common benign tumour of the sinonasal tract (Liu *et al.*, 2010). Osteomas of jaw bones are quite rare (Núñez-Gi, 2009).

The location of osteomas of the jaws is usually in close proximity to areas of muscle attachment, suggesting that muscle traction may play a role in its development (Sayan *et al.*, 2002). Small osteomas are harmless and can be left untreated (Meher *et al.*, 2004).

Case reports of osteoma with imaging findings are extremely scarce. The location of osteoma in the occipital region is extremely rare and only one case has been reported so far (Meher *et al.*, 2004). Multiple osteomas of head and neck region can be presented as an isolated finding or may be associated with Gardner's syndrome. The case report presented here is an isolated case with multiple involvements of the craniofacial region.

## Case report

A 21-year-old female patient presented with a complaint of swelling in the forehead and posterior to the left ear since she was 3 years old. The swelling gradually increased over the years to the present size. Patient gave no history of change in vision, headache or hearing impairment. The swelling was asymptomatic and her past medical history was not significant. There was no history of similar findings within the family.

On extra oral examination, there was facial asymmetry due to the presence of the swelling in the forehead and the left lower border of the mandible (Fig. 1). Maxillary anterior prognathism and lip trap were also observed. A similar kind of swelling was observed near the left mastoid region (Fig. 2) and occipital region. The patient had incompetent lips. Intra oral examination revealed wide maxillary arch with high arched palate, malaligned teeth and gingival overgrowth seen in relation to the right upper and lower posterior teeth.

Blood investigations revealed all components within the normal range. Endoscopy and colonoscopy were performed to rule out Gardner's syndrome. Orthopantomograph revealed impacted 33, retained deciduous 73, altered tooth morphology in relation to 16, 17, 47 and 48, pulp stone in relation to 47 and radiopaque was observed posterior to 17 (Fig. 3). A lateral cephalogram revealed bony enlargement in the frontal region with

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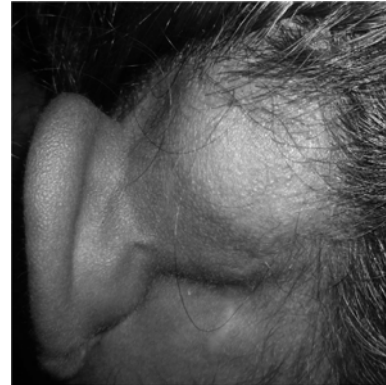
scalloping in the interior of the lesion. A similar enlargement was noticed near the chin (Fig. 4). CT revealed presence of hyperdense areas in relation to left mastoid area, occipital and predominantly in the left frontoethmoidal region (Fig. 5 and 6). Three-dimensional reconstruction revealed osteoma in frontal, mastoid, occipital and in the symphyseal area (Fig. 7, 8 and 9).

Patient was merely concerned with

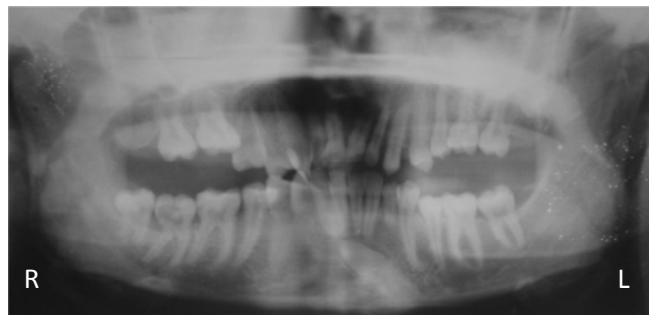
aesthetics and so only cranioplasty of the frontal region was carried out and the specimen was examined histopathologically which revealed presence of dense lamellae with organized haversian canals and the intratrabecular stroma showed osteoblasts, fibroblasts, and giant cells, with no hematopoietic cells (Figure 10). Patient was followed for a period of six months and no recurrence was found (Figure 11).



**Figure 1** Clinical photograph of the patient showing swelling on the forehead region and inferior border of the mandible.



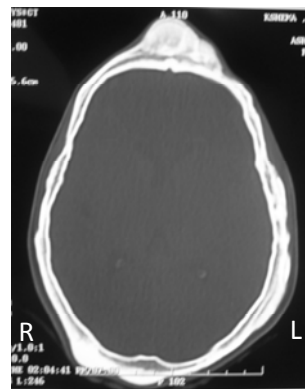
**Figure 2** Clinical photograph showing swelling in the left post auricular region.



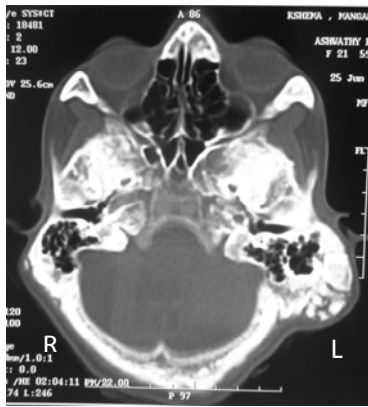
**Figure 3** Orthopantomogram showing presence of impacted canine in the mandibular anterior region.



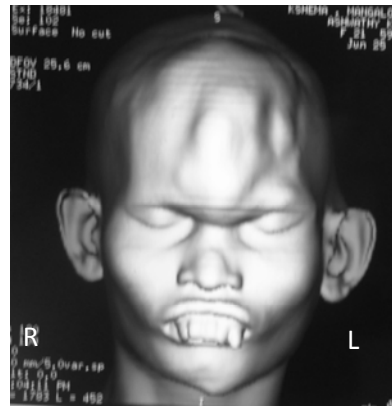
**Figure 4** Lateral cephalogram showing osteomas in the frontal sinus and protruded maxilla.



**Figure 5** CT section showing multiple hyperdense areas in the frontal and occipital region.



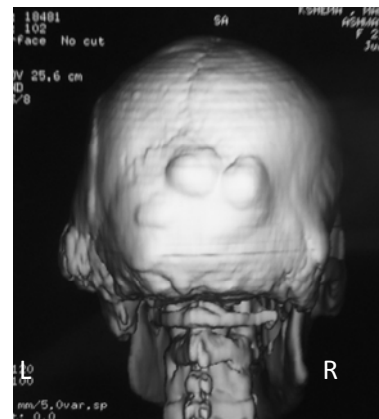
**Figure 6** CT section showing hyperdense areas in the left mastoid area.



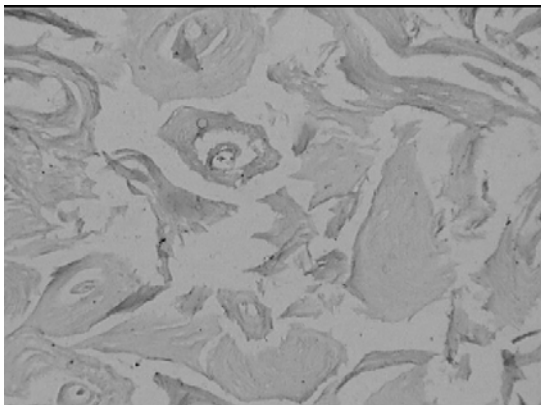
**Figure 7** Three dimensional reconstruction showing frontal osteoma, maxillary prognathism and symphyseal osteoma in the mental region.



**Figure 8** Three dimensional reconstruction showing left mastoid osteoma.



**Figure 9** Three-dimensional reconstruction showing multiple occipital osteomas.



**Figure 10** Histopathologic image of the osteoma (H&E 10X) showing dense lamellae with organized Haversian canals and the intratrabecular stroma contains osteoblasts, fibroblasts, and giant cells, with no hematopoietic cells.



**Figure 11** The photograph showing the profile view of the patient after the surgical correction of frontal osteoma.

## Discussion

Osteomas are common in the frontoethmoid region and defined as a benign, circumscribed, slow-growing bony tumor of the mastoid (Stuart, 1940). The incidence of osteomas is highest in frontal followed by, ethmoid and maxillary sinuses (Meher *et al.*, 2004). Osteomas of the cranial vault are rare (González *et al.*, 2006).

The mastoid osteoma can arise from any part of the temporal bone. By 2006, there were about 150 cases of mastoid osteomas reported in the literature (González *et al.*, 2006). The aetiology is not clear and various theories have been suggested which include trauma, infection and hereditary factors (Schwartz, 1961). Stuart suggested that a pituitary dysfunction might influence this condition (Stuart, 1940).

Temporal osteomas are found rarely before puberty and are most common in females, as seen in the case presented here, whereas middle ear osteomas are common in males (Hornigold *et al.*, 2003). The case presented here had involvement of the fronto- ethmoidal region, mastoid, occipital region, mandible and the maxilla.

Microscopically, osteomas are characterized by dense lamellae with organized haversian canals. The intratrabecular stroma contains osteoblasts, fibroblasts, and giant cells, with no hematopoietic cells (Fenton *et al.*, 1996). Similar histopathological features were seen in our case.

Imaging of the osteomas can be achieved by traditional radiography or CT scan. The use of CT scanning with 3-D reconstruction makes it possible to achieve a better visualization and more precise localization (Bodner *et al.*, 1994). On a CT scan osteomas may present as demarcated and hyperdense outgrowths as seen in this report. Axial and coronal scans together demonstrate the exact dimension of osteomas.

The lesions are usually asymptomatic but sometimes can present with symptoms like headache, invasion and deformity of the orbit, pneumocephalus with possible rhinorrhea and meningitis, and, rarely, abscess formation (Haddad *et al.*, 1997). These symptoms were not reported in the case presented here.

Although this tumour has a considerable incidence, there are few reports with large samples on this subject (Larrea-Oyarbide *et*

*al.*, 2008). The case presented here had multiple site involvement which is usually seen in Gardener's syndrome. Gardener's syndrome is characterized by polyps of the colon, multiple osteomas, and multiple impacted or unerupted teeth, with skin and soft tissue tumours. Cutaneous findings include epidermoid cysts, desmoid tumours, and other benign tumours (Madani and Madani, 2007). As the osteomas develop before the colorectal polyposis, early recognition of the syndrome may be a life saving event in some cases (Woldenberg *et al.*, 2005). As such, the only reported finding for this patient was one impacted canine in the mandible.

The treatment for osteomas is surgical resection and recurrence is rare after complete removal. In our case surgical correction was carried out in the frontal region.

## Conclusion

An isolated case of multiple osteomas of head and neck region is a rare phenomenon and was reported in this case report.

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