



Giant Osteoma of the Mandible: Report of a Rare Case with Review of Literature

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Abstract Osteoma is a slow growing, asymptomatic, benign bony tumor composed of compact and cancellous bones. Central, peripheral, and extra skeletal osteomas are the three types based on the site of origin. They are mostly observed on routine radiographic screening, mostly in the paranasal sinuses. Gnathic involvement is an uncommon occurrence, and if present, mandibular involvement is more frequently seen. Mostly, osteomas are small asymptomatic lesions and very rarely they become symptomatic and acquire larger size. Multiple osteomas are a feature of Gardner's syndrome; however, solitary osteomas are non-syndromic. Oral health professional may be the first to diagnose Gardner's syndrome as the osteomas may be initial manifestation of the disorder. Treatment protocol of osteomas varies based on the associated signs and symptoms. Small, asymptomatic cases are treated conservatively by periodic clinical and radiographic evaluation. However, larger, symptomatic lesions require surgical intervention. Herby, reporting an unusual case of Giant peripheral osteoma of the mandible. Our case is unique in few aspects because of its unusually large size (5 × 4 cm) and involvement of lingual aspect of the mandible in the region of sublingual fossa, with compression of the floor of mouth.

Keywords Gardner's syndrome · Giant osteoma · Mandible · Osteoma

Introduction

An osteoma refers to a painless, gradually progressing benign bone neoplasm typified by proliferation of mature spongy and/or compact bone [1].

Osteomas can be delineated into three types based on the site of origin: Central, originates from the bony endosteum; peripheral, protrudes from the bone surface; or heterotopic, originates apart from bone in soft tissue (muscles) [2, 3]. Peripheral osteomas (Pos) exhibits centrifugal expansion from the periosteum, while central osteomas exhibit centripetal growth from the endosteum [4].

However, the recent World Health Organization (WHO) nomenclature delineates only two types—central and surface osteomas [2].

Osteomas are primarily confined to the craniofacial structures, with a site predilection for paranasal sinuses (primarily frontal sinus followed by ethmoid and maxillary sinus) [5]. Gnathic skeletal involvement is, however, rarely seen [6].

Generally, peripheral osteomas manifest as an asymptomatic slow growing bony mass, usually small in size [7]. On the contrary, large osteomas may result in facial disfigurement, and may displace and injure the adjoining framework [7, 8].

Individuals with multiple osteomas should be thoroughly appraised to rule out Gardner's syndrome. As Gardner's syndrome exhibit a genetic predisposition, new osteomas may appear after a few months or years. Hence, a periodic review every 4–6 months is essential for patients with Gardner's syndrome [9–11].

Oral physicians should always bear in mind that most osteomas are an incidental finding on regular radiographic screening, hence, oral physicians should be familiar with

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the varied presenting manifestation of this clinical entity [12].

The treatment protocol relies on a myriad of factors—size, site, extent of the lesion along with its proliferative pattern and accompanying manifestations. Asymptomatic osteomas are generally treated conservatively, and periodic imaging is recommended to observe the growth rate. However, surgical intervention is reserved for symptomatic cases [13, 14].

Here, we report a rare case of a giant solitary peripheral osteoma of mandible arising from the lingual aspect of mandibular body and occupying the floor of mouth. A brief review on the published literature of giant osteoma of mandible has also been presented.

Case Report

A 65-year-old female reported to the out-patient Department of Oral Medicine and Radiology, Saveetha Dental College and Hospitals, Chennai with a chief complaint of swelling beneath the right side of the tongue for the past 10 years. History reveals that the patient noticed an asymptomatic swelling beneath the right side of tongue 10 years back. The swelling showed slow progressive growth to attain the present size; however, there has been no increase in the size for the last 1–2 years. Negative history of facial trauma was elicited. Family history was unremarkable for gastrointestinal symptoms, and no similar swelling was noticed in any other part of the body. Dental history was positive for multiple tooth extractions.

Physical examination was unremarkable with no constitutional symptoms. Extraoral examination revealed a

firm, non-tender swelling in the right sub mental region with non-palpable regional lymph nodes (Fig. 1a). Intraoral examination revealed a solitary, pinkish-red, lobulated swelling of size 5 cm anteroposteriorly and 4 cm mesiodistally in the floor of the mouth on the right side. The swelling extended anteriorly to the symphysis region, posteriorly up to the distal aspect of the second molar, medially up to the base of tongue causing deviation of tongue postero-laterally, and laterally up to the lingual aspect of mandibular body and revealed a cranial growth. The swelling caused lateral and posterior displacement of tongue, thus, interfering with speech and mastication. The surface of swelling was smooth with no inflammatory component. The mouth opening was within the normal range with no alteration in lip, chin, and tongue sensations. The oral hygiene was poor with multiple missing teeth, generalized tooth mobility and cervical abrasion. On palpation, the inspeactory findings such as size, site, shape, surface, and extent were confirmed. Palpatory findings suggested that the swelling was bony hard in consistency, non-tender, fixed to the mandible and does not yield on pressure. The borders of the swelling were well defined and merged with the base of the tongue and floor of the mouth (Fig. 1b, c).

Mandibular occlusal radiograph and Lateral oblique radiograph revealed well-defined radio opacity in the right side of mandible, not surrounded by radiolucent rim (Fig. 2a, b) OPG showed a well delineated radio opaque lesion in right mandible. The radio-opacity extended antero-posteriorly from the right symphysis region to the distal aspect of second molar, and mesio-distally from the lingual aspect of right side of body of mandible to the floor of the mouth. The radio-opaque mass was not surrounded

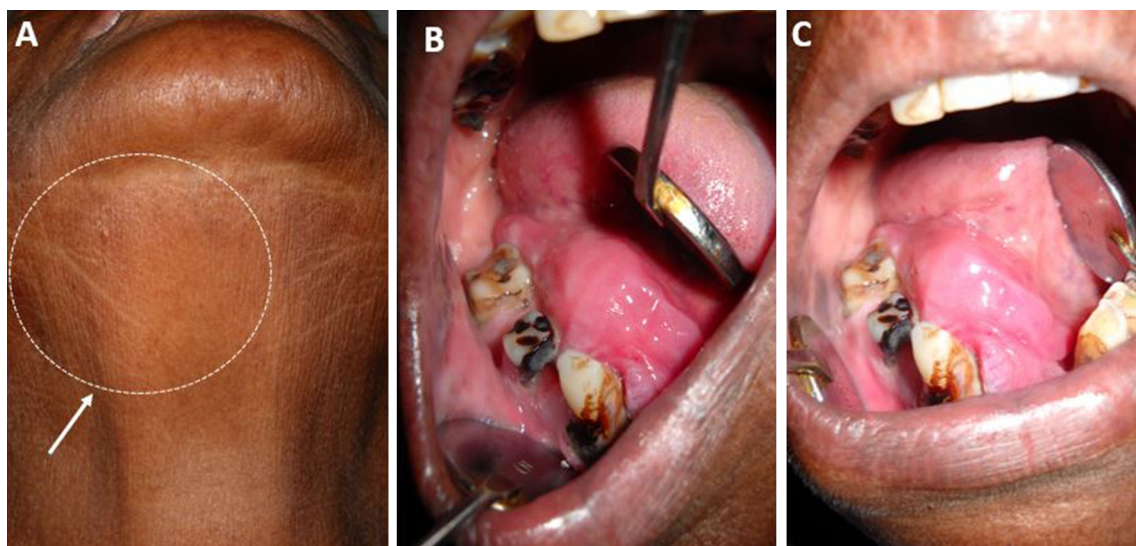


Fig. 1 a Extraoral swelling in the submental region. b, c Intraoral extent of the swelling in the floor of mouth

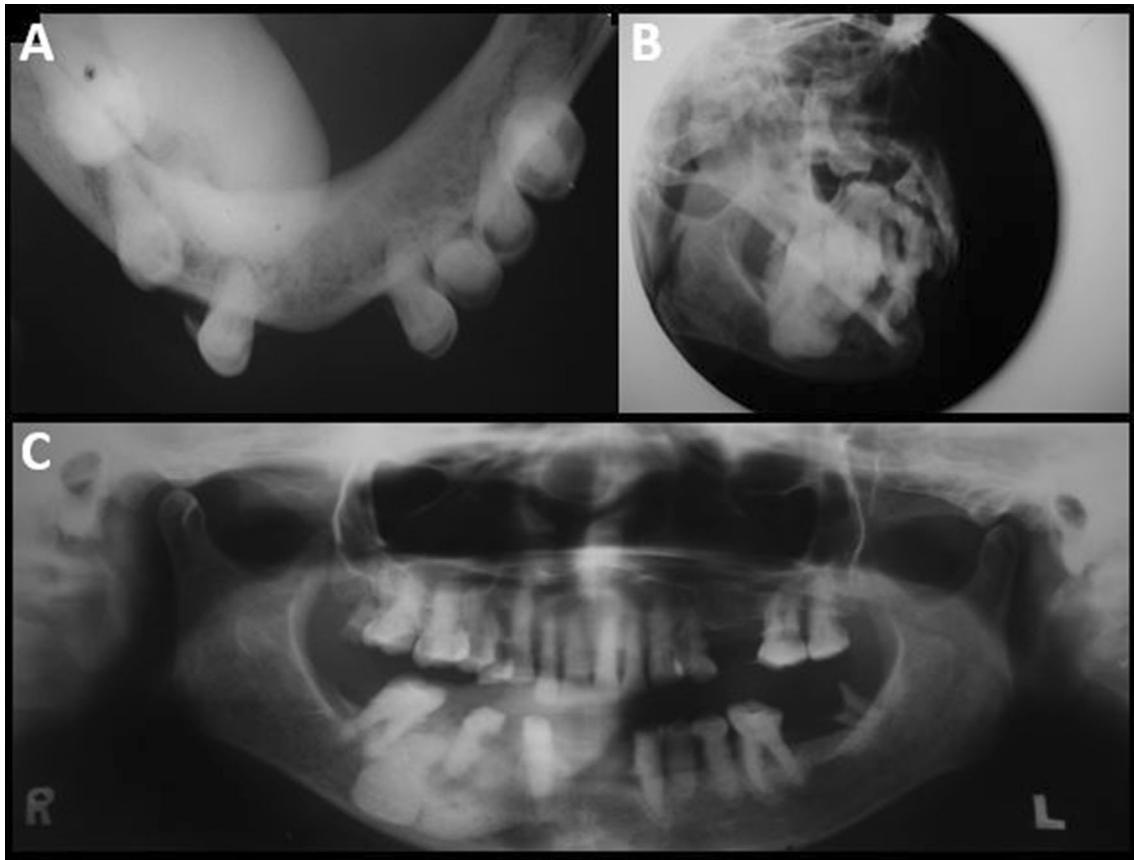


Fig. 2 Mandibular cross-sectional occlusal radiograph, lateral oblique & OPG depicts a well-defined homogeneous radiopaque mass in the right mandibular body

by a radiolucent rim. OPG also revealed multiple missing teeth, generalized interdental bone loss with ill-defined periapical pathology in relation to medially displaced second premolar (Fig. 2c).

CT scan revealed a moderately sized pedunculated, hyper dense bone lesion of size $3.5 \times 3.4 \times 1.8$ cm arising from the lingual aspect of body of mandible on the right side in the region of sublingual fossa. The lesion was seen compressing the adjacent floor of mouth (Fig. 3a, b).

Fig. 3 Axial and coronal CT shows the extent of homogeneous radiopaque mass



Based on the history, clinical examination, and radiological features, provisional diagnosis of peripheral osteoma was considered, and Exostosis, odontoma, osteoid osteoma, osteoblastoma and ossifying fibroma were placed in the list of differential diagnosis.

Comprehensive blood parameters, liver and kidney function tests were in the normal range. The patient was instructed about the nature of the lesion and advised surgical intervention. After a written consent, mass was

surgically resected in toto raising the mucoperiosteal flap. Healing was uneventful and no complications were noted (Fig. 4a). Histopathological examination of the decalcified tissue revealed mature compact lamellar bone trabeculae with minimal marrow spaces, and prominent osteoblastic activity, features consistent with a diagnosis of compact-type peripheral osteoma (Fig. 4b, c).

However, the patient was lost for further follow up.

Discussion

According to the recent WHO nomenclature, osteomas are classified as a benign neoplasm of well differentiated mature bone tissue [15], and Jaffe [16] first described the condition [15, 17].

A solitary peripheral osteoma of the gnathic skeleton is an uncommon occurrence, with a prevalence of less than 1%. However, mandibular involvement is more frequently seen, in contrast to the maxilla [6, 18].

Mandibular osteomas exhibit a site predilection for the angle and lower border of the body of mandible (mostly in association with the buccal cortical plate). Involvement of the lingual aspect of mandibular body, such as in our case, is an uncommon occurrence [6, 12, 19, 20].

Etiology of osteomas is still controversial. Some researchers have proposed the neoplastic or developmental basis of the entity [5, 9, 10, 18].

The embryological or developmental theory advocates development of osteomas from sutures between bones of diverse embryological origin. However, this theory could not be substantiated as osteomas usually progress in adults and not during childhood or adolescence [7, 20].

The persistent growth pattern exhibited by osteomas during adulthood is the most distinctive characteristic, thus, demarcating these lesions from other bony exostoses, and advocating the neoplastic theory of origin [10, 20]. However, other researchers deny the neoplastic origin because of the very slow growth rate.

Donohué Cornejo et al. [21] demonstrated the neoplastic activity by scintigraphy in an active giant osteoma of the mandible.

Furthermore, Bulut et al. [7], Kaplan et al. [10], Woldenberg et al. [22] proposed that osteomas may develop as a reactive lesion due to trauma, infection, or muscle activity.

Minor trauma may result in subperiosteal edema or bleeding and the muscle activity could locally elevate the periosteum, thus, triggering an osteoproliferative response that could be sustained by the unremitting muscle traction [10, 15, 20].

Most peripheral osteomas occur in proximity with muscle insertions (masseter, medial pterygoid, and temporalis); thus, supporting the reactive genesis secondary to muscle traction [6, 9, 10, 18, 22].

The inflammatory theory proposes that chronic infection of paranasal sinuses triggers proliferation of periosteum associated bone cells. However, it is still debatable to ascertain it is the infection or osteoma that develop earlier. Moreover, this theory fails to elucidate pathogenesis of osteomas in other sites [7].

The lesion in our patient was in contiguity to mandibular teeth with chronic periodontitis. The trigger factor for osteoma formation in our case could possibly be attributed to the poor oral hygiene leading to multiple tooth extractions.

Unilateral, well-circumscribed, sessile/pedunculated, mushroom shaped masses of size 1.5–4 cm and exhibiting a slow, persistent growth pattern generally summarizes the clinical features of peripheral osteomas [7, 20, 23, 24]. The term “gigantiform or huge osteoma” may be designated to large sized untreated osteomas (> 3 cm in size or 110gm in weight) [17, 20, 25].

Giant osteoma of the mandible is an exceptionally rare entity, and to the best of our knowledge (after thorough PUBMED, Scopus and Google scholar data base), only 30 cases of giant peripheral mandibular osteomas have been reported so far in the literature (Table 1).

Our case is unique in few aspects because of its unusually large size (5 × 4 cm) and involvement of

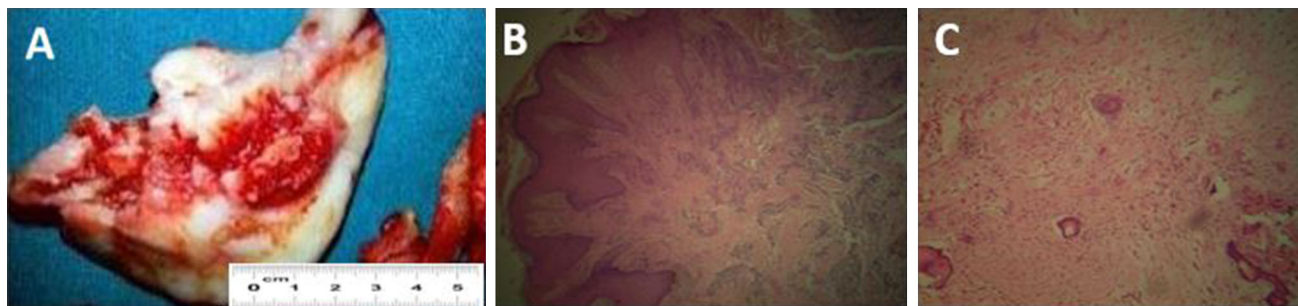


Fig. 4 a Macroscopic resected specimen. b Osteomatous element seen beneath the surface epithelium with interspersed thin fibrous layer in between. H&E 100 × (c)

Table 1 Depicting documented Giant mandibular osteoma cases

S.no	Author	Year	AGE/sex	Type	Symptom	Size (cm)	Location of lesion
1	Richards HE et al.	1986	NA	NA	NA	NA	Genial tubercle region
2	Swanson KS et al.	1992	22/M	NA	Asymptomatic	NA	Right posterior mandible
3	Kerckhaert et al.	2005	53/F	Central	Dysphagia	NA	Right body of mandible
4	Young et al.	2006	60/M	Peripheral	Dyspnea	8.7	Lingual surface of Left mandibular angle; extended from submandibular space to infra temporal space
5	Kalu et al.	2007	50/F	Peripheral	Dysphagia	NA	Lingual aspect of Right posterior body of mandible
6	Mittal A et al.	2008	30/M	Peripheral	swelling	4X4.1X4cm	Left submandibular region, angle & Lower border
7	Bulut E et al.	2010	37/F	Peripheral	Swelling	3 × 3 cm	Buccal aspect of Left mandibular body
8	Donohué Cornejo et al.	2010	7/F	NA	Asymptomatic swelling	3 × 3	Left angle and ramus of mandible
9	Kaya GS et al.	2010	55/F	NA	Asymptomatic mass	NA	Lingual aspect of left mandibular body
10	Gouder et al.	2011	29/M	NA	Asymptomatic swelling	3	Body of Mandible
11	Saati S et al.	2011	26/F	Compact	Dull aching swelling	3.9 × 4.2 cm	Lingual aspect of left mandibular body
12	Almeida et al.	2011	52/F	NA	Swelling over the preauricular region & TMJ pain	NA	Right Condyle & glenoid fossa
13	Kachewar et al.	2012	50/F	Peripheral	Swelling	10.8 × 4.0x4.6 cm	Body, ramus, angle & lower border of mandible
14	Soni S et al.	2012	35/F	Peripheral	Dyspnea and dysphagia	6 × 3 cm	Lingual aspect of right mandibular body
15	Soni S et al.	2012	60/M	Peripheral	Swelling	5 × 3 cm	Right mandibular angle; submandibular region & ascending ramus
16	Tarsitano et al.	2013	41/M	NA	Sleep apnoea	NA	Right mandibular body and angle
17	Sadeghi HM et al.	2015	53/M	Central	Swelling	9.5 × 8 cm	Left mandibular body, Angle & ramus
18	Gawande et al.	2015	45/M	Osteoma	Swelling	5 × 4 cm	submandibular area; buccal and lingual surface of the body, ramus, angle and the inferior border of right mandible
19	Shetty SK et al.	2015	16/F	Cancellous osteoma	Swelling & recurrent pain	4 × 5 cm	Left mandibular Ramus and angle region
20	De Souza PD et al.	2015	41/F	Peripheral	Painless Swelling	3 × 2 cm	Left submandibular region
21	Pulkit K et al.	2016	40/F	Peripheral	Dysphagia	6 × 3 cm	Lingual aspect of left mandibular body
22	Ohashi et al.	2016	65/F	Central	Incidental discovery on CT	7.3 × 4.1x3.7 cm	Right mandible
23	Nilesh K et al.	2016	75/F	Central	swelling	5 × 3 cm	Mandibular alveolus
24	Sagar P et al.	2017	30/M	NA	swelling	7X5cm	Lower border of left mandible
25	Xin Xu et al.	2017	43/M	NA	Limited mouth opening	3 × 2 cm	Medial aspect of left condyle

Table 1 continued

S.no	Author	Year	AGE/sex	Type	Symptom	Size (cm)	Location of lesion
26	Bozkurt P et al.	2018	43/F	NA	Swelling; Pain & difficulty in mastication	NA	Buccal aspect of mandibular body
27	Heitz C	2018	NA	Peripheral	NA	NA	Condyle
28	Saha A et al.	2019	10/M	Central	Swelling	3.0 × 3.6×3.9 cm (CT scan dimensions)	Left body of mandible
29	Sachin Ram G et al.	2019	40/M	NA	Swelling	4 × 3 cm	Medial aspect of left mandibular angle
30	Demircan S et al.	2020	17/M	NA	Swelling; asymmetry & dysphagia	4.5 × 3.5×3cm	Left mandibular Ramus

lingual aspect of the mandible in the region of sublingual fossa, with compression of the floor of mouth.

Osteomas may be seen in any age group. However, most cases are seen in the 30–50-year age range, with a slight predilection to occur in males than in females [2, 3, 5, 8, 9, 12, 19, 26, 27].

However, other researchers documented that osteoma occur mostly in the younger age group with no gender predilection [7, 28, 29]. Our patient was 65-year-old female who noticed the swelling some 10 years back.

Generally, most POs are asymptomatic and are incidental finding on regular clinical and radiographic assessment. Gross facial asymmetry and swelling with hindered functions (difficulty in chewing, swallowing, breathing, and restricted mandibular movements) may be seen in larger lesions [7, 17, 19, 25, 28, 30]. Mandibular osteomas may also present alteration in bite and dentition [29].

The present case revealed a huge, solitary bony mass in the floor of mouth, and interfered with speech and mastication.

Various imaging techniques like occlusal radiographs, orthopantomogram (OPG), Waters projection, computerized tomography (CT), Cone beam computer tomography (CBCT), and magnetic resonance imaging (MRI) play an essential role in periodic evaluation of POs [15]. CT is an excellent imaging aid for diagnosis, anatomical location, and size determination of the lesion [8, 11, 17, 23, 28, 30].

Radiographically, POs appear as ovoid, well-defined, radiopaque, sessile/pedunculated lesions of size 1–4 cm in diameter, with a density comparable to that of normal bone [3, 4, 7, 9, 11, 18, 22, 25, 30]

These features are in coherence to our case report. These lesions usually do not damage the adjoining bony structures as reported in our case [3].

POs appear as well-circumscribed, lobulated, round-ovoid, sessile/pedunculated, mushroom shaped hyperdense lesions. Sessile osteomas are frequently connected to the bony cortex with a wide stalk, in contrast to the pedunculated lesions which have a narrow bony contact [7].

However, pedunculated lesions are not always apparent radiographically, hence, an incisional biopsy is imperative for a confirmatory diagnosis [28].

Microscopically, POs presents in 2 forms. Compact osteoma appears as solid lamellar bone mass with minimal marrow tissues; and Cancellous osteoma presents as trabeculae of mature lamellar bone trabeculae with interspersed fibrous or fatty marrow [2, 3, 9, 28, 31]. Generally, POs exhibit a pronounced osteoblastic activity. Most gnathic POs are microscopically compact osteomas, in coherence with the present case.

Exostoses, osteoid osteoma, osteoblastoma, peripheral ossifying fibroma, complex odontoma and late-stage central ossifying fibroma, must be contemplated in the differential diagnosis of POs.

Exostoses such as tori are common bony outgrowths (usually on the labial/buccal region of the alveolar bone), and are considered hamartomatous, developmental or reactive lesions, rather than true neoplasms. Varied growth pattern with growth cessation after puberty characterizes these lesions [3, 7, 19, 24, 26, 28].

Osteoblastoma and osteoid osteoma are rapidly growing, painful lesions chiefly affecting younger age group. Pain due to osteoblastoma decreases with aspirin, in contrast to osteoid osteoma, where pain diminution with aspirin does not occur [11, 19, 32].

Our patient reported an independent slow growth pattern and was symptom free during the entire course of the lesion, thus, ruling out the possibility of exostosis, osteoid osteoma and osteoblastoma.

Ossifying fibroma is a reactive lesion affecting the teenagers and young individuals. Reactive exuberant enlargement of the interdental papillae in maxillary anterior region, with a well-circumscribed, radiopaque mass not invading the bony cortex usually constitute the clinical and radiographic features [11, 24].

Central ossifying fibroma usually manifest as a well-defined radiopacity surrounded by a thin radiolucent line separating it from the adjoining bone. A sclerotic border

may be present in the bone next to the lesion [19, 32]. Thin radiolucent line is the key distinguishing radiographic feature with POs [24].

Complex odontoma appears as a well delineated radiopaque mass encircled by a thin radiolucency, with a density greater than the abutting bone. [6, 19, 32].

Our case presented as a well-defined lobulated radiopaque mass without a radiolucent rim, thus ruling out the possibility of odontoma and ossifying fibroma.

Multiple osteomas in a patient may alert the physician for a possible Gardner's syndrome. Multiple intestinal polyps (strong risk predisposition for malignant transformation), multiple skeletal osteomas/enostosis, and multiple impacted supernumerary teeth constitute the cardinal features of this syndrome. Osteomas usually precede the development of intestinal polyps, and oral physician may be the first to diagnose this condition, thus, ameliorating the prognosis of the disorder [3, 5, 7, 8, 10].

Our patient did not report any gastrointestinal symptoms or dental anomaly. Family history and clinical examination was also inconclusive for Gardner's Syndrome.

Varied treatment approach exists for POs. Smaller asymptomatic lesions generally require periodic radiographic evaluation, whereas, surgical excision is reserved for lesions which are large, disfiguring, symptomatic lesions exhibiting expeditious growth pattern and jeopardizing the functional ability [5, 7, 8, 12, 18, 19, 33].

The tumor mass was surgically resected in our patient, and healing occurred uneventfully. Recurrence of POs is an exceptionally uncommon occurrence. Only 3 cases of recurrence have been documented till date [34–36]. However, there has been no evidence of malignant transformation of POs [3, 5, 6, 11, 17, 18, 22, 28].

Conclusion

Osteomas are benign bone neoplasms exhibiting slow persistent growth pattern. Periodic clinical and radiographic assessment plays a key role in the management of osteomas. Oral health professional should be familiar with the presentation, as multiple osteomas are a feature of Gardner's syndrome. CT is the imaging modality of choice. Recurrence after surgical intervention is extremely rare.

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Declarations

Conflict of interest The author declares that they have no conflict of interest.

Human Participants and/or Animals The patient and the attendants were informed about the nature of disease and treatment protocol.

Informed Consent The patient was informed about the nature of disease and treatment protocol. Written informed consent was taken from patient.

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