Revisiting Peripheral Osteoma of the Mandible with Case Series and Review of Literature

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Abstract Osteomas are benign lesions composed of mature compact and cancellous bone usually arising from craniofacial region. They can be central or peripheral, solitary or multiple. We are presenting few cases of giant peripheral osteoma's of the mandible along with review literature of other jaw lesions with their diagnostic features and differential diagnosis. The purpose of presenting this paper is to evaluate radiopaque lesions of the jaw which require proper clinical and histological evaluation to reach a final diagnosis.

Keywords Peripheral osteoma · Radiopaque lesions

Introduction

Osteoma is a benign neoplasm in which apposition of newly formed bone creates a tumor mass [1]. The majority of cases occur in the craniofacial bones most frequently in paranasal sinuses and jaw bones, although rare cases in other bones and in soft tissues such as muscle have been documented [1]. Two variants of osteoma are recognized which differ in the origin in relation to cortical plates: Peripheral (periosteal) osteomas develop as masses attached to cortical plates and central

osteomas arise from endosteal bone surface [1, 2]. Peripheral osteoma of craniofacial region occurs most frequently in the paranasal sinuses. Other locations include external auditory canal, orbits, temporal bone and pterygoid process [3, 4]. Clinically, peripheral osteoma appears as a unilateral, sessile or pedunculated, well circumscribed, mushroom like mass larger from 10 to 40 mm in diameter. There is no predilection for age, sex [3–7]. Solitary central osteomas of the jaws seems to be very rare with only six such cases reported in English language literature since 1955 [8–14]. Central osteoma are difficult to diagnose and has to be differentiated with other similar lesions of the jaws like central ossifying fibroma, condensing osteitis, dense bone island and osteoblastoma, as well as cementoblastoma and odontoma in cases occurring within tooth bearing areas. Multiple osteomas of jaws are a hallmark of gardener syndrome (familial adenomatous polyposis) an autosomal dominant diseases caused by a mutation in the APC tumor suppressor gene [15]. In this syndrome patient has multiple osteomas along with multiple premalignant colorectal adenomas, which if left untreated progress to colorectal carcinoma by middle age. Histologically osteomas may be of two types: (1) Compact or “ivory” and (2) Cancellous, trabecular or spongy. The compact osteomas comprises of dense, compact bone with a few marrow spaces and with only a few osteons. The cancellous osteoma is characterized by bony trabeculae and a fibro fatty marrow enclosing osteoblast and with an architecture resembling mature bone [4, 5, 16–19]. The aim of this paper is to Present clinical, radiological and histopathological feature of few cases of peripheral osteoma of mandible occurring in the anterior and posterior mandibular region with the review of literature and their surgical treatment.

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Case Reports

Case I

A 35 year old female (Fig. 1) patient reported to our institute with a chief complaint of a large stony hard mass in her oral cavity with difficulty in eating food and breathing since last 2 years. On palpation the lesion was stony hard, non-tender, firmly adherent to the underlying lingual aspect of the mandible. Overlying mucosa was intact and appeared healthy.

Lingual cortical expansion was present the lesion was approximately $6 \times 3 \text{ cm}^2$ in size extending from left canine region to the right ascending ramal region of mandible. There was gross deviation of the tongue to the contralateral side with restricted movement. The tongue was positioned posteriorly on the unaffected side causing mild respiratory discomfort to the patient. A peculiar finding in the case was that the entire mass on closure of mouth was almost touching the hard palate. No evidence of neurological deficits of the tongue or the mandible were present. Routine investigations, X-ray mandible and CT scan neck was advised. The scanned images were suggestive of a large dense radiopaque mass adherent to the lingual aspect of symphysis menti, body and ascending rami. There was no evidence of compression of great vessels of the neck. A segmental resection of mandible was performed from left parasymphyseal region of mandible to the right mid rami. Post operative recovery and healing was uneventful (Fig. 2). Histological specimen was suggestive of a peripheral osteoma.

Fig. 1 Patient with a clinical diagnosis of peripheral osteoma

Fig. 2 Postoperative view

Case II

A 60 years old male presented to our institute with a chief complaint of swelling in the right side of the jaw since 1 year with mild pain over swelling since 2 months. The swelling was initially small in size which progressively increased in size from last 3 months. There was pain over swelling which was dull aching and of non-radiating type. There was no history of swelling over buccal aspect of posterior mandible, trismus, or any other ear, nose, or throat complaints. On examination patient had hard fixed, non-tender swelling about $5 \times 3 \text{ cm}^2$ in size on right angle region of mandible extending up to right submandibular region and involving part of ascending ramus of mandible. Overlying skin appeared to be normal. Intraoral lingual cortex of mandible was not expanded. There was no mucosal perforation and normal overlying mucosa and gingiva appeared normal. X-ray mandible showed well defined oval lobulated radiopaque lesion at right angle of mandible. A CT scan (Fig. 3) mandible showed a well defined bony lesion consisting of compact bone with no trabecular pattern arising from inner aspect of right side of mandible with no soft tissue component and causing mild displacement of great vessels of neck. CT scan findings were suggestive of Giant ivory osteoma arising from angle of right mandible. After consultation with Oral and Maxillofacial consultant excision of tumor with reconstruction of mandibular defect with titanium reconstruction plates was planned. Patient was taken for surgery. During surgery tumor was found to involve whole of the base of angle of mandible along with a part of ascending ramus of mandible. So preservation of cortex of mandible was not possible. A segmental resection of posterior right mandible was planned intraoperatively. An L shaped 3 mm titanium reconstruction plate (Fig. 4) was fixed to retain the anatomy of the resected mass. Post operative follow up of the

patient was done every 6 months for one and a half years. No complications were reported.

Case III

A 38 year old healthy female reported to our institute with a chief complaint of a progressively enlarging asymptomatic hard growth in lower front tooth region towards the tongue since last 1 year. On palpation the mass was stony hard, non-tender sessile and oval in shape. The overlying mucosa was intact with no signs of inflammation. The mass was around $3 \times 1.5 \text{ cm}^2$ in size with extension from lower left side premolar area to right side lower molar region. The growth had a sublingual extension into the floor of the mouth, but the growth was not invasive or infiltrative in nature. Tongue deviation was minimal and there was no evidence of paresthesia over tongue or entire mandibular region. A CT scan was advised which reported a non-enhancing well margined oval shaped lesion with no bony invasion or soft tissue destruction. The radiographic diagnosis of a peripheral osteoma of left anterior mandible with extension to right body region of mandible was made. Segmental resection of the mass from body to body region of mandible was performed and a L shaped titanium reconstruction plate was fixed for aesthetic and functional considerations. A post operative 3D cone beam CT was advised (Fig. 5). Histopathologically the sections were suggestive of cortical lamellar bone containing osteon with haversian canals consistent with Osteoma. The histopathologic diagnosis confirmed the clinical diagnosis of peripheral osteoma. The patient presented for postoperative visits and suture removal a week later, there were no postoperative complications. Prosthetic rehabilitation with a removable partial denture was also performed after 6 month. None of the three cases cited in the study have reported any recurrence till date.

Fig. 3 Axial CT image of case II depicting a well defined bony lesion of right angle of the mandible

Fig. 4 A L shaped titanium reconstruction plate in situ

Fig. 5 Cone beam CT image of Case III

Discussion

Osteomas of the jaws are rare osteogenic lesions considered to be true neoplasm [1]. However some peripheral osteomas may be reactive rather than neoplasm associated with trauma [2]. Peripheral osteomas typically present as mushroom shaped, hard radiopaque masses which are often pedunculated but may also have a broad base by which they are attached to the cortical plates. Osteomas displays a very slow growth rate however they will continue to grow slowly if left untreated. Since osteomas of the skull can be part of Gardner's syndrome, this possibility has to be excluded. This syndrome is based upon the presence of numerous adenomatous colorectal polyps, soft and hard tissue abnormalities and congenital hypertrophy of the retinal pigment [20]. Inheritance is autosomal dominant. The incidence of this syndrome varies from 1:8,300 to 1:16,000 live births [20]. The prevalence of osteomas in the normal population is about 4 %. One of the study conducted with familial adenomatous polyposis syndromes reported percentage of osteoma ranging from 57 to 82 % [21]. Osteomas often go undetected, since they are usually asymptomatic unless incidentally noted on routine

Table 1 Clinical and radiographic characteristics of the cases from the literature and the three new cases

Source	Age	M/F	Jaw	Location	Radiodensity	Border definition	Size (cm)	Expansion	Displacement	Symptoms
1 Hitchin et al [8]	47	M	Mandible	Premolar/molar	RO	WD irregular	N1	Yes	Edentulous	Pain, lymphadenitis sensory deficit
2 Khosla [9]	13	M	Mandible	Molar	Mixed	WD	3 cm	Yes	Yes	Pain carious adjacent teeth
3 Rajayogeswaranav [10]	69	F	Maxilla	Anterior	RO	WD	1.2 cm	No	Edentulous area	Sharp/dull pain
4 Friz [11]	22	F	Mandible	Molar/within alveolar Canal	RO	WD	1 cm	No	No	Pain burning spontaneous fasciculation of lip
5 Zielinska Kazamierska [12]	20	F	Mandible	Premolar/molar	RO	WD irregular	7 × 2.5 cm	Yes	Displacement & impaction	Pain, multiple fistula
6 Firat et al [13]	15	M	Maxilla and sinus	Premolar	RO	WD irregular	Approx 3 cm	No	Displacement	Mild pain, nasal & impaction\obstruction
7 Kaplan et al. [29]									Alerted sensation	
Case 1	46	M	Maxilla	Premolar/molar	RO	WD	1.7 × 13	Yes	No	None
Case 2	52	F	Mandible	Premolar/molar	RO	WD	2 × 2	Yes	No	None
Case 3	67	F	Mandible	Canine/premolar	RO	WD	1 × 1	Yes	No	None
Case 4	57	M	Mandible	Premolar	RO	WD	1 × 2	Slight	Yes	None
8 S Soni.A.Bhargava 2012 (Present Report)										
Case 1	35	F	Mandible	Canine/molar	RO	WD	6 × 3 cm	Yes	Yes	Deviation of tongue & mild airway obstruction
Case 2	60	M	Madible	Angle	RO	WD	5 × 3 cm	No	No	None
Case 3	38	F	Mandible	Premolar/molar	RO	WD	3 × 1.5 cm	Mild	No	None

RO radiopaque, WD well defined, NI unavailable information

Table 2 Differential diagnoses central radiopaque lesion of the jaws

Age	Age (Decades)	M/F	Radiodensity/borders	Expansion	Displacement/resorption	Symptoms	Histopathology
Central ossifying Fibroma	3rd–4th	F > M	RL/Mixed RO WD RL rim	Yes	Possible	No	Cellular fibrous tissue Bone trabeculae
Cementoblastoma	2nd–3rd	No predilection	RO fused to root RL rim	Yes	Possible	No	Cellular/acellular cementum
Odontoma (compound)	2nd	No predilection	Amorphous RO mass RL rim	Yes	Often	No	Unorganized enamel/dentin/cementum
Idiopathic osteosclerosis	Any	No predilection	RO Smooth or scalloped margin	No	No	No	Thickened trabeculae reduced marrow fibrovascular spaces
Osteoblastoma	2nd	M > F	RL/RO well/poorly defined	Possible	Possible	Pain Discomfort Common	Irregular trabeculae Vascular Stroma
End-stage focal osseous dysplasia	3rd–6th	F ≫ M	Mixed or RO irregular WD borders	No	Uncommon	No	Cellular fibrous tissue bone and cementum
Central osteoma	2nd–5th	M > F	RO/WD	Yes	Possible	No	Cancellous/compact bone

RL radiolucent, RO radiopaque, WD well defined borders

radiographic survey. Osteomas are often noted in the sixth decade, but report range from 16 to 74 years [22].

We have enumerated various cases of osteomas reported till date in literature in Table 1. Differential diagnosis of osteomas include peripheral ossifying fibroma, exostoses, sessile osteochondroma, periosteal osteoblastoma, osteoid osteoma, and periosteal osteoblastoma, parosteal osteosarcoma [23]. A differential diagnosis of various radiopaque lesions of the jaws has been enumerated in Table 2 for ready reference.

Peripheral ossifying fibroma is a reactive focal overgrowth that occurs predominantly in the anterior portion of maxilla and is characterized histologically by a prominent collagenous highly cellular stroma. Peripheral ossifying fibroma presents as a radiopaque mass but does not intrude into osseous cortex [24]. Central ossifying fibroma is a neoplasm which is well defined radiolucent to radiopaque. It may mature with time into a totally radiopaque mass. Characteristically it is surrounded by a thin radiolucent rim. COF may cause expansion and root displacement or resorption. The main feature to differentiate it from osteoma radiographically is the radiolucent rim [25].

Exostosis are hamartomas with predilection for the lingual [torus mandibularis] and buccal regions of the mandible, midline of hard palate [torus palatinus], buccal and palatal region of maxilla [26]. Usually exostosis stops growing after puberty [23]. In tooth bearing area cementoblastomas, should be included in the differential diagnosis of central osteomas. Cementoblastoma is a neoplasm which typically presents as a radiopaque mass with well defined borders which may cause root resorption and expansion or delayed eruption in the rare cases associated with primary dentition [28, 29]. The pathognomonic characteristic of cementoblastoma is its fusion both macroscopically and microscopically with the root of a tooth. From a histologic point of view it may be difficult to differentiate between bone and osteocementum; therefore the diagnosis is based upon the direct continuation between involved root surface and the lesion which help to rule osteoma as a possibility [29]. In sessile osteochondroma, the cortex of the lesion merges imperceptibly with the cortex of the host bone. It is composed of areas of endochondral ossified calcified cartilage and a fatty or haemopoietic marrow in the trabecular spaces. In contrast osteoid osteomas is a lesion of rapid growth, frequently painful and microscopically features highly vascular cellular tissue containing osteoid tissue [4, 5]. Periosteal osteoblastoma presents as a round or ovoid heterogenous mass attached to the cortex, is rapidly growing and painful. Microscopically it is composed of trabeculae of woven bone with osteoblast and osteoclast [5].

Odontomas also may present as densely radiopaque mass with well defined borders in tooth bearing areas of

jaws. Complex odontomas may present as a relatively uniform solid radiodensity, and therefore only these cases could be considered in the differential diagnosis of central osteomas. They can cause displacement, expansion and delayed eruption. Odontomas are thought to cease growing at the age at which normal odontogenesis is completed [29].

Parosteal osteosarcoma presents as a lobulated, sclerotic mass, homogenous and heterogenous in density with more radiolucent areas peripherally. An incomplete cleft between the lesion and adjacent cortex is occasionally present. Microscopical examination revealed stromas of woven lamellar bone in a heavily collagenized stroma [23].

Osteblastoma should also be considered in the differential diagnosis of radiopaque lesions of the jaws. They occur predominantly on the left side of the posterior mandible and are associated with pain, tenderness and discomfort. Microscopically osteblastoma is characterized by a proliferation of osteoblast forming bone trabeculae set in a vascularized fibrous connective tissue stroma, often with osteoblastic rimming [27]. Osteomas are devoid of the background of vascularized fibrous connective tissue. This is the main differentiating feature [29]. Review of the english literature demonstrated peripheral osteomas of the jaws in only 69 well documented cases. According to the study of Aline et al. [23] which demonstrates the details of six cases in the maxilla and 63 cases in the mandible. In the most frequent site affected in upper jaw was the alveolar process followed by hard palate. The ratio female to male was 2:1 as per the study the mandibular cases published in the literature suggested that most common site is body (posterior region is more common than anterior), followed by condyle, angle, ascending ramus, coronoid process and sigmoid notch. However, in the anterior region of the mandible, the female:male ratio was 3:1. Aline et al. 2005 study also suggest the mean age of 36.5 years (9–85 year) with 28.5 year. range (9–65) for the anterior region.

The histopathologic aforesaid type ratio found according to the study was cancellous:compact of 1:1.35. However, cancellous osteomas are reported as rare in the literature [17].

In our study of three cases the patient's age was within 9–85 years. Posterior region of the mandible was more commonly involved. Both male and female were equally affected in the mandible.

Conclusion

In our study we have presented few cases of peripheral osteomas which most commonly occur in lower jaws, occurs at the surface of cortical bone and can be sessile or

pedunculated. These radiopaque lesions of jaws require proper clinical, radiological and histopathological evaluation for proper diagnosis and treatment planning. Due to the fact that similar characteristics are presented with other radiopaque lesions of the jaws proper histological evaluation is necessary to reach the final diagnosis.

Conflict of interest The author and the co-author hereby declare that we do not have any financial relationship with any organization and that our research was not sponsored. We do not have any conflict of interest.

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