CASE REPORT

Peripheral osteoma of the body of mandible

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SUMMARY

Osteoma is a benign osteogenic neoplasm microscopically consisting of proliferation of cancellous or compact bone. Peripheral osteomas (PO) arise from the periosteum and are quite uncommon in the jaw bones. POs of mandible are considered as rare entity and very few cases have been reported in the literature. The pathogenesis of PO is unclear. Some investigators consider it a true neoplasm, while others believe it as a developmental anomaly, a reactive mechanism due to trauma or infection. The purpose of this article is to present the clinical, radiographic, surgical and histological features of a solitary peripheral osteoma of the mandible in a 43-year-old woman and to review the literature for PO located in the mandible.

BACKGROUND

Osteoma is a benign, mesenchymal, slow-growing osteogenic tumour commonly occurring in the craniofacial bones, characterised by the proliferation of compact and/or cancellous bone. Solitary osteomas may be classified as: peripheral (paraosteal, periosteal or exophytic) when arising from the periosteum, central (endosteal), when arising from the endosteum and extraskeletal (the so-called osseous choristoma) when arising in soft tissue. Multiple osteomas may be associated with Gardner's syndrome. A peripheral osteoma (PO) occurs most frequently in the paranasal sinuses. Other locations include the orbital wall, temporal bone, pterygoid processes and external ear canal and are rarely seen in the mandible.¹⁻⁶ A review of the literature revealed only 16 well-documented cases.⁷ The purpose of this article is to present a case of peripheral osteoma of the body of mandible and to review features of case/s of PO in the craniofacial region described in the literature.

CASE PRESENTATION

A 43-year-old woman was referred to the department of oral and maxillofacial surgery by a general

dentist, for evaluation of a hard swelling in the left side of the lower jaw of approximately 2 years duration. A distinct facial asymmetry was observed extraorally with slight elevation in the body of the mandible, 1 cm above the lower border (figure 1). Intraoral examination revealed no lesion and all other findings including the status of the dentition were within the normal. The lesion was asymptomatic and hard in consistency. The patient was in good health with no history of previous facial trauma, surgery or any additional contributory medical factors.

INVESTIGATIONS

A panoramic radiograph demonstrated a well circumscribed, round to oval-shaped radiopaque mass between roots of the left lower first and second molars (figure 2). CT showed a pedunculated, cauliflower-like growth protruding from the surface of the compact bone (figure 3A). A three-dimensional CT also revealed a lobulated bony growth in the same area (figure 3B).

DIFFERENTIAL DIAGNOSIS

Clinical and radiographic differential diagnosis was osteoma and osteochondroma.

TREATMENT

An excisional biopsy was performed by making an extraoral trapezoid incision with periosteal elevation and the mass was exposed (figure 4). The lesion from its base was removed completely using rotary instruments and a chisel. The surgical specimen was submitted for the microscopic examination (figure 5). Histologically, the specimen showed dense compact bone with little fibrous connective tissue stroma at the periphery and no evidence of any inflammation or malignancy (figure 6). No haversian systems typical of normal bone structure were found (figure 7). A diagnosis of 'peripheral osteoma of the body of the

Figure 1 Clinical photographs (lateral and frontal views) showing the swelling in the mandibular body region.

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Figure 2 Orthopantomograph showing a round to oval-shaped radiopaque mass seen between left mandibular first and second molars.

mandible' was made from the clinical, radiographic and histopathological findings.

OUTCOME AND FOLLOW-UP

The patient is on regular periodic follow-ups and has remained disease free from the past 1- year postsurgery.

DISCUSSION

Osteoma is a benign osteogenic lesion characterised by proliferation of compact or cancellous bone. It can be central, peripheral or extraskelatal. Central osteomas arise from the endosteum, peripheral osteoma from the periosteum and extra-skeletal soft tissue osteoma within a muscle.⁸

PO of the jaw bones is quite rare. Sayan *et al*² reported finding 22.85% of the lesions in the mandible and 14.28% in the maxilla in their study. However, reports by Kaplan *et al*⁷ and Woldenberg *et al*⁴ showed that 81.3% and 64% of cases, respectively, were occurred in the mandible. The systematic review of the literature of PO of lower jaws showed 63 cases published as various case reports and reviews. PubMed and MEDLINE search was done using the keywords osteoma, peripheral osteoma, bone tumours and osteoma of jaws for the review of literature. The recent review was published by Johann *et al*.⁹ In the present article, an additional 19 cases have been included after the recent review.^{4 10–19} The summary of clinical details such as age, gender, site of the previous cases of PO of lower jaw in the literature up to date is enlisted in table 1.

A search of the literature identified a total of 82 well-documented cases of solitary peripheral osteoma of the lower jaws till 2012. Of 82 cases, 41.5% of cases are from the body of the mandible, 23.2% from the condyle, 14.6% from the angle,

11% from the ascending ramus, 8.5% from the coronoid process and 1.2% from the sigmoid notch. Very rarely, as in the present case, the lesions are located on the buccal aspect of the body of the mandible. The age range was 9–85 years in the earlier review. In the present article, the age range is between 13 and 75 years. The previous review showed an equal predilection towards men and women for the occurrence of PO of the mandible. In contrast, our review showed 19 additional case reports of which 14 cases in women and 5 cases in men since the last review published by Johann *et al*.⁹ The pathogenesis of PO is still controversial.^{20 21} The exact aetiology and pathogenesis of peripheral osteoma is unknown. Neoplastic and reactive causes have been suggested as possible aetiological factors. Some researchers have considered that osteomas arise from either embryological cartilaginous rests or persistent embryological periosteum.²² Others consider them to be true neoplasms.^{7 20} POs are probably not neoplastic in nature, because in the majority of cases their growth potential and growth rate seem to be limited;⁷ although trauma, inflammatory or infectious processes are the causes commonly cited in the literature.^{7 8 20} Certain researchers have reported that many POs are located on the lower border of the mandible, and it is possible that muscle traction plays a role in the development of peripheral osteomas.^{4 11} The possibility that PO may be a reaction to trauma was first suggested by Thoma.²³ If one accepts the reactive pathogenesis of PO, then the term osteoma seems to be inappropriate because it implies a benign neoplastic process. Few considered that a term that better reflects the non-neoplastic nature of PO is 'parosteal osseous hyperplasia'.⁷ Minor trauma, not likely to be remembered by a patient years later may cause subperiosteal oedema or bleeding, and the muscle traction could locally elevate the periosteum. This can initiate an osteogenic reaction that could be preserved by the continuous muscle traction.⁸ In the present case, the patient did not have any history of trauma or infection to the lower jaw.

Clinically, POs are usually asymptomatic, sometimes a firm or hard swelling is noted. The swelling is usually painless and can be discovered during routine clinical and radiographic examination. Sometimes, depending on the location and size of the lesion, it may cause swelling, facial asymmetry and functional impairment.^{4 5 11 22–26} Multiple osteomas of the jaw with supernumerary impacted teeth, dermoid cysts, multiple colon polyps and desmoid tumours indicate Gardner's syndrome.²⁷ Gardner's syndrome must be excluded in patients with osteoma.²⁸ In our case no such features were observed.

On radiological imaging, a peripheral osteoma of the mandible appears as well-circumscribed, round to oval, mushroom-like radiopaque mass with distinct borders.^{9 11 29} Traditional

Figure 3 CT shows a pedunculated, cauliflower-like growth and a lobulated bony growth in the same area seen from three-dimensional CT.

Figure 4 The exposed mass after surgical incision.

radiographic techniques are generally sufficient to diagnose these lesions.³⁰ Panoramic radiography or CT is sometimes used for imaging. However, reports in the literature suggest CT as the best imaging modality for determining the location and real extension of the lesion.^{4 5 20 24 25 31} Three-dimensional CT are also used and found to be more helpful in showing the relationship between the tumour and the adjacent structures than the routine radiographs.²⁰

Histologically, osteomas are composed of a normal-appearing, dense mass of lamellar bone. Although they contain osteoblasts, fibroblasts and giant cells in intertrabecular stroma, haemopoietic cells are rarely observed. Osteoma presents a variable amount of osteoblastic and osteoclastic activity.³² On the basis of microscopic findings, osteoma can be referred as compact osteoma containing marrow tissue or cancellous osteoma showing trabeculae of mature lamellar bone with intervening fatty or fibrous marrow. The compact osteoma usually has a sessile base, normal-appearing dense bone with minimal marrow spaces, and occasional Haversian canals. The cancellous osteoma is usually pedunculated and resembles the bone of origin, containing trabeculae of bone and fibrofatty marrow with osteoblasts. The surface can be irregular or smooth, with cortical bone at the margin. The size of the different types of osteoma ranges from

Figure 5 The macroscopic appearance of the lesion submitted for histopathology after removal.

Figure 6 Photomicrograph showing tumour mass consisting of dense compact bone with little fibrous connective tissue stroma at the periphery (H&E stain, ×100 magnification).

several millimetres to several centimetres. However, part of the lesion may be in bone, masking the true size.³³

Clinically, peripheral osteoma should be differentiated from several pathologies, such as exostoses, osteblastoma, osteoid osteoma, late-stage central ossifying fibroma or complex odontoma.¹³ Exostoses are bony excrescences that occur on the buccal aspect of alveolar bone. These lesions are of reactive or developmental origin and are not thought to be true neoplasms.³⁴ Usually exostoses stop growing after puberty.³⁵ In contrast, osteoid osteomas are lesions with rapid growth, are frequently painful and microscopically feature highly vascular cellular tissue containing osteoid tissue.^{2 33} Periosteal osteoblastoma presents as a round or ovoid heterogeneous mass attached to the cortex, is rapidly growing and painful. Microscopically it is composed of trabeculae of woven bone with osteoblasts and osteoclasts.³⁶ Peripheral ossifying fibroma is a reactive focal overgrowth that occurs, predominantly, in the anterior portion of the maxilla and is characterised histologically by a prominent collagenous highly cellular stroma. The peripheral ossifying fibroma presents as a radiopaque mass, but does not intrude into the osseous cortex.³⁷

Recurrence of peripheral osteoma after removal is extremely rare^{4 33} and there are no reports of malignant transformation.⁴

Figure 7 Photomicrograph showing normal bony structure without Haversian system (H&E stain, ×400 magnification).

Table 1 Summary of clinical data of peripheral osteoma cases of the mandible published in the literature

Serial No	Age*	Gender	Site	Authors
1	19	F	Condyle	Mancini <i>et al</i> ¹⁰
2	31	F	Body of mandible	Woldenberg <i>et al</i> ⁴
3	40	M	Angle	Woldenberg <i>et al</i> ⁴
4	16	F	Ascending ramus	Woldenberg <i>et al</i> ⁴
5	17	M	Body of mandible	Woldenberg <i>et al</i> ⁴
6	13	F	Body of mandible	Woldenberg <i>et al</i> ⁴
7	79	F	Body of mandible	Woldenberg <i>et al</i> ⁴
8	46	F	Condyle	Woldenberg <i>et al</i> ⁴
9	26	F	Body of mandible	Woldenberg <i>et al</i> ⁴
10	13	M	Body of mandible	Woldenberg <i>et al</i> ⁴
11	50	F	Body of mandible	Ogburke <i>et al</i> ¹¹
12	75	F	Ascending ramus	Cincik <i>et al</i> ¹²
13	37	F	Body of mandible	Bulut <i>et al</i> ¹³
14	25	M	Body of mandible	Rodriguez <i>et al</i> ¹⁴
15	34	F	Body of mandible	Shakya ¹⁵
16	45	M	Condyle	Almeida ¹⁶
17	35	F	Body of mandible	Mubeen <i>et al</i> ¹⁷
18	15	F	Angle	Singhal <i>et al</i> ¹⁸
19	33	F	Angle	De Franca <i>et al</i> ¹⁹

Source: Modified and updated from Johann *et al.*⁹

*Age in years.

F, female; M, male.

Learning points

- Although peripheral osteomas of jaws are rare, this case demonstrates that if neglected they can enlarge slowly and considerably.
- Cosmetic disfigurement and functional impairment are main reasons of patients opting for the treatment. The postsurgical follow-up should comprise periodic clinical and radiographic studies.
- Patients with osteoma associated with impacted or supernumerary teeth should be evaluated for the possible Gardner's syndrome.
- However, it is very essential to rule out Gardner's syndrome as a solitary osteoma could be the earliest sign of indication of this syndrome.

Competing interests None.

Patient consent Obtained.

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