REVIEW PAPER



Intraosseous Leiomyoma of the Mandible: A Case Report of the Rare Entity and Review of Literature

Kirti Chaudhry Dutt¹ · Sukhvinder Bindra² · Meenakshi Awana³ · Manjit Talwar³ · Gurvanit Lehl³

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Abstract

Introduction Leiomyoma is a rare tumor of smooth muscle origin with a very low incidence in the maxillofacial region. Intraosseous occurrence of oral leiomyoma is even rare with involvement of mandible followed by maxilla. Aim The purpose of this paper is to present a case of intraosseous leiomyoma of the left mandibular angle region with the review of literature describing this rare entity. The paper also highlights the need to include this entity in differential diagnoses of jaw lesions.

Material and Method An extensive search of literature was carried out on the Medline-Pubmed and Google Scholar database using the keywords leiomyoma, maxilla, mandible, oral and palate to thoroughly search and collect all the reported cases of intraosseous leiomyoma.

Result To the best of our knowledge till date only 22 cases of intraosseous leiomyoma have been reported in the

maxillomandibular region we represent the 23rd case of the intraosseous leiomyoma in a 36 year old male patient.

Conclusion Though uncommon but it is known to occur in the jaws therefore intraosseous leiomyoma should be included in the list of differential diagnoses of radiolucent lesion of mandible and maxilla.

Keywords Intraosseous · Mandible · Maxilla

Introduction

Leiomyoma is a benign tumor of smooth muscle origin, which is usually associated with uterus, GIT and skin [1]. Oral cavity is devoid of smooth muscles except in blood vessel wall thus expectedly this tumor is quiet rare in oral and maxillofacial region with reported incidence of 0.065 %. This accounts for 0.42 % of all soft-tissue neoplasms in the oral cavity. When present in the oral cavity common sites of involvement are lips (27.46 %), tongue (18.30 %), cheeks and palate (15.49 %), gingiva (8.45 %) and mandible [2]. Intraosseous occurrence of oral leiomyoma is even rarer with involvement of mandible followed by maxilla.

Farman et al. [3] in a study found that out of 7748 smooth muscle tumors only five cases (0.06 %) were present in the oral cavity. Leiomyoma was first described by Virchow in 1854 and first case of oral leiomyoma was reported in 1884 by Blanc [4]. Since then many cases of oral leiomyomas have been reported but majority of these are confined to soft tissues. Until date, only 23 cases of intraosseous leiomyoma in orofacial region have been reported.

In this paper we are presenting a new case of mandibular leiomyoma along with a systematic review of the English literature.

Kirti Chaudhry Dutt sukhikirtiomfs@gmail.com

Meenakshi Awana sukhvinder151275@gmail.com

Manjit Talwar talwarmanjit7@yahoo.in

Gurvanit Lehl gvlehl@yahoo.co.uk

- Department of Dentistry, All India Institute of Medical Sciences, Jodhpur 342005, India
- Department of Oral and Maxillofacial Surgery, Army College of Dental Sciences, Secunderabad, India
- Department of Dentistry, Government Medical College and Hospital, Sec – 32, Chandigarh 160030, India



Sukhvinder Bindra sukhvinder151275@gmail.com; sukhikirtiomfs@gmail.com



Fig. 1 An OPG depicting a radiolucency around periapical region of the left mandibular third molar



Fig. 2 The excised tissue specimen of approximately 5 cm in size

Case Report

A 36 year old male patient presented to Department of Dentistry, Government Medical College and Hospital, Sec—32, Chandigarh with a swelling on left mandibular angle region since about 3 months. Swelling was painless with no other signs of inflammation. Medical history was unremarkable. Patient did not report any paresthesia. On dental examination patient was found to have pericoronitis in relation to left mandibular third molar without any sinus or tooth mobility.

The orthopantamogram revealed a well-defined unilocular radiolucency (Fig. 1) around 2–3 cm in size in the periapical region of the left third molar, which was not associated with any resorption of roots or displacement of the tooth. A provisional diagnosis of odontogenic cyst or tumor was considered and thus enucleation with extraction



Fig. 3 The excised tissue specimen of approximately 5 cm in size

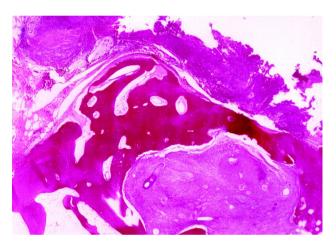


Fig. 4 The histopathological picture demonstrating the solid variety of intraossoeus Leiomyoma

of third molar was planned under local anesthesia through intraoral approach. The neurovascular bundle was preserved during surgery and no paresthesia was observed post operatively.

Excised tissue (Figs. 2, 3) was sent for histopathological examination (Fig. 4), which showed fibrous connective tissue stroma along with bony trabeculae. Under higher magnification the connective tissue appeared to be arranged in interlacing bundles of whorls and fasicular patterns. The cells of the tissue were elongated, having spindle shaped nuclei showing tapered ends and open chromatin arrangement. This was typical of smooth muscle fibers. The rest of the connective tissue showed dispersed collagen bundles and irregular bony trabeculae with osteocytes in lacunae. Scattered areas of osteoid tissue and few interspersed endothelial lined blood vessels were also seen.

These histopathologic features were conclusive of soild variety of intraosseous leiomyoma.



Table 1 Summary of total cases of leiomyoma in maxilla or mandible found in literature

no Rhatigan et al. [6 Coldblatt et al. [7	INCIDITE S	rigo alla	Drecentation	Rone	Size of lesion	Root	Cortical	Padiology	Becurrence	Histological
Ω 0		sex		involved	10101 10 0710	resorption	plate	(9)		diagnosis
9	hatigan et al. [6]	71 years/ F	Painful swelling. Teeth mobility since 1.5 years	Maxilla— posterior area	1 cm	Yes	Perforation	Radiolucent area with sclerotic margins	Follow up time 2 years—no recurrence	Cellular leiomyoma
	oldblatt et al. [7]	3.5 years/ F	Bleeding gums	Mandible— left posterior area	2 cm × 2.5 cm	DNF	Expansion	Radiolucency	Follow up time 25 months—no recurrence	Leiomyoma
3 Mech [8]	Mechlin et al. [8]	21 years/ F	Gradual swelling and mild pain since 2 years	Maxilla—right posterior area	8 cm × 7 cm	DNF	Perforation	Radiolucent lesion destroying lateral and inferior maxillary sinus wall and alveolar ridge	DNF	Leiomyoma
4 White [9]	White et al. [9]	61 years/ M	DNF	Mandible— anterior and posterior area	4 cm	DNF	DNF	Multilocular radiolucency	Follow up time of 6 years—no recurrence	Angiomyoma
5 White [9]	White et al. [9]	69 years/ M	Not reported	Mandible- anterior and posterior area	More than 2 cm	Not reported	Not reported	Multilocular radiolocency	Followup time of 6 years—no recurrence	Angiomyoma
6 Mc M et a	Mc Millian et al. [10]	54 years/ M	Painless swelling since 2 months	Mandible— posterior area	>2 cm	Yes	Perforation	Multilocular radiolucency	Follow up time 9 month—no recurrence	Angiomyoma
7 Raffin [11]	Raffini et al. [11]	8 months	Painless swelling since 2 months	Mandible— right posterior area	4 cm	DNF	Perforation	Unilocular radiolucency	Follow up time 12 months— no recurrence	Solid leiomyoma
8 Bhatt [12]	Bhatt et al. [12]	25 years/ M	Painful swelling	Mandible— posterior area	0.5 cm ⁻¹ cm	Yes	Expansion	III defined radiolucency	DNF	Angiomyoma
9 MacLeod et al. [1	lacLeod et al. [13]	12 years/ M	Painless swelling since 3 weeks	Mandible— left Posterior area	<2 cm	DNF	Expansion	Unilocular radiolucency	No recurrence	Solid leiomyoma
10 Burkes Jr et al. [14	urkes Jr et al. [14]	36 years/ M	None. Radiographic finding	Mandible— posterior area	1.5 cm × 2.5 cm	DNF	DNF	Well defined unilocular radiolucency with sclerotic borders	Follow up time 1 year—no recurrence	Angiomyoma



Solid lieomyoma Solid leiomyoma Solid leiomyoma Solid leiomyoma Solid leiomyoma Angiomyoma Recurrence not Angiomyoma leiomyoma Histological diagnosis Vascular DNF DNF Follow up time Follow up time Followup time-Follow up time Follow up time Follow up time Follow up time no recurrence Follw up time 11 years—no no recurrence 2 years—no 2 years—no 5 years—no 2 years—no 16 months— 40 months-1 year. No recurrence recurrence recurrence recurrence recurrence mentioned Recurrence DNF sinus with wall distruction Opacification of maxillary Radiolucency surrounding Large radiolucency—type with sclerotic margins Well defined unilocular Unilocular radiolucency Unilocular radiolucency Unilocular radiolucency root of lateral incisor Radiolucency with unerupted canine not mentioned radiolucency Radiology DNF DNF mentioned) mentioned Perforation Perforation Perforation Perforation Perforation DNF (not Cortical DNF DNF DNF mentioned mentioned reported resorption None None None None Root DNF DNF Not Not Yes $1.4 \times 1.3 \times 1$ cm Not Size of lesion $6.5 \times 5 \text{ cm}$ $2 \times 1.5 \text{ cm}$ 9.5 and 0.8 cm 2 cm 4 cm 4 cm 2 cm DNF DNF Maxilla- right and anterior and anterior anterior and Mandibular– third molan Mandible— Mandible— Mandibular Mandible— Mandible— Mandibular Mandible— Retromolar posterior posterior posterior posterior posterior posterior Mandible anterior nvolved area area area area area left left swelling since **Footh mobility** Asymptomatic. Asymptomatic. Asymptomatic radiographic radiographic radiographic Asympomatic Aymptomatic right nasal Presentation Progressive 6 months 3 months stuffiness 6 months Routine swelling swelling Routine finding Painless Painless finding Painless since since 13 years/ 24 years/ 75 years/ 57 years/ 27 years/ F Age and sex 24 years/ 12 years/ 49 years 9 years/ M male Labruna et al. Konstantinos et al. [23] Loyola et al. Brooks et al. Liang et al. [19] Suresh et al. Katau et al. et al. [20] Koca et al. References Luis et al. Fable 1 continued Bertolini [18] [21] [24] [15] Serial no 12 13 4 15 16 18 19 20 17



angioleiomyoma Solid leiomyoma Histological diagnosis Solid Follow up time Follow up time Follow up time no recurrence No recurrence 18 months recurrence 2 yrs—no Recurrence 1 year Radiolucency with sclerotic Radioleucency Radioleucency Radiology Perforation Cortical None None resorption None None $10 \times 10 \times 5 \text{ mm}$ Size of lesion 1 cm right body Mandible— (posterior posterior Mandible involved area) Palate swelling since Pink to slightly anterior palate red nodule in discomfort Presentation 2 months Painless 36 years/ Age and sex 35 years/ 9 years/ M Reddy et al. [26] Present case et al. [25] References Grossman Serial 22 23

 Fable 1
 continued

DNF Data not found

Material and Method

We systematically searched the Medline-Pubmed and Google Scholar database using the keywords leiomyoma, maxilla, mandible, oral and palate. But our search was not just limited to these terms only. We checked the references of these articles for other cases and thoroughly studied them. In total, we found 23 cases of intraosseous oral leiomyomas. In some case reports leiomyoma initially originated in soft tissue and progressed to involve the bone as reported by Yamomoto et al. [5] while in many cases authors have not distinguished between intraosseous and soft tissue oral leiomyoma. The latter have not been included in our review. The clinical and radiographic findings of these cases are shown in Table 1.

Discussion

Leiomyoma is defined by the World Health Organization as a "circumscribed benign, often cutaneous tumor composed of intersecting bundles of mature smooth muscle cells." [2]. Leiomyoma of the mandible is very uncommon. The rarity of intraosseous leiomyoma could be well explained by paucity of smooth muscle cells in the bone where the origin is only likely from tunica media of blood vessel wall [27] or some heterotopic embryonal tissue [28]. Whereas in oral cavity other sources like circumvallate papillae [29], ductus lingualis [30] for the tongue and aberrant arrectores pilorum muscles for the cheek have also been additionally suggested. In the present case radiographic and surgical findings suggested an intraosseous origin and could be from the smooth muscle of vascular walls or heterotopic embryonic tissues.

It could be seen at any age. In our review of 23 patients, mean age of occurrence is found to be 39.15 years, which includes a range from 8 months to 75 years. Our patient is 36 years old and our findings are in agreement with the findings of Reddy BVR et al. [26].

In this analysis of 23 patients, it was observed that males were affected more than females in the ratio of 2:1. The male predominance is concurrent with the literature [31].

Clinical presentation may vary from being very asymptomatic to painful swelling with or without cortical destruction. Non-specific pain may be the main manifestation of intraosseous leiomyoma [31]. In the cases reviewed, eight patients had asymptomatic swelling, four had mild pain with swelling. Five patients have not reported with any clinical sign and symptom. Three patients experienced nasal stuffiness, bleeding gums [6] and tooth mobility [14].

Most of the lesions were found in mandibular posterior followed by mandibular anterior region. Only two cases



have been reported in maxillary posterior area. This is in accordance with the review of Liang et al. [19].

Radiographically, intraosseous leiomyoma presents itself as radiolucency, which may be unilocular or multilocular with either ill or well-defined sclerotic borders. Most of the cases in our review were of unilocular variety. The small size of the mass and the rim of sclerosis suggest a slow rate of growth consistent with benign tumor. Lesions may lead to cortical destruction/expansion or even root resorption or tooth displacement depending on the nature and location of the lesion. As intraosseous leiomyoma, being a rare entity, is usually not considered in the radiographic differential diagnosis. More likely diagnostic considerations for this radiographic presentation include central giant cell lesion, ameloblastoma, myxoma, traumatic bone cyst, haemangioma, neurofibroma and sarcoma [19]. In our case radiolucency was seen around apical area of third molar and we also thought this to be an odontogenic tumor or cyst and planned the treatment accordingly.

Histologically, leiomyoma has abundant spindle shaped cells arranged in whorled or interlaced fascicular pattern. The major cytological findings are elongated nuclei having fusiform or blunt ends and typical perinuclear vacuoles.

Leiomyomas have been classified histopathologically into three groups by WHO: leiomyoma (solid leiomyoma); angiomyoma (vascular leiomyoma); leiomyoblastoma (epitheloid leiomyoma) [32]. Solid leiomyoma is composed of a proliferation of spindle-shaped cells arranged in thick intersecting bundles. Mitoses are very rarely seen [32].

Angiomyomas have tortuous mass of thick walled blood vessels between intertwining bundles of smooth muscle. The epitheloid leiomyoma has rounded or a polygonal cell with a clear zone around the nucleus and the cytoplasm is acidophilic. Duhig and Ayer [33] stated that vascular leiomyoma is a step in the transformation from haemangioma to solid leiomyoma through a process of continuous proliferation of smooth muscles.

Svane [34] in reviewing 114 cases of leiomyoma found that 74 % were vascular leiomyoma, 25 % were solid and only 1 % were leiomyoblastomas. Baden [31] reviewed 142 cases of oral leiomyoma and found 67 % to be vascular, 31.7 % to be solid leiomyoma and only 1.3 % to be leiomyoblastoma. In our review also most of the patients including our case reported to have solid variety of leiomyoma and eight patients had angiomyoma.

It would appear that introsseous leiomyomas histologically are not predominantly vascular, as are the soft tissue oral leiomyomas.

Histopathological abundance of spindle shaped cells adds other benign lesions like myofibroma, hemangiopericytoma, neurofibroma, neurilemmoma and schwannoma to its differential diagnoses. Masson's trichome stain may be used to differentiate leiomyoma from neurofibroma or neurilemmoma as it stains smooth muscle fibres red and collagen fibres green or blue. Damm and Neville [35] recommend Mallory's phosphotungstic acid hematoxylin as the most suitable stain for demonstrating myofibrils. Immunohistochemical and electron microscopic observations are of great help in achieving an accurate diagnosis.

It is very important to differentiate leiomyoma from its malignant counterpart leiomyosarcoma. It is found that larger size, rapid increase in size; cortical destruction are some of the indicators of malignancy. However intraosseous leiomyoma have been reported to have larger size than their soft tissue counterparts. Tumour doubling time is also used as an indicator for malignancy. In gastric leiomyomas tumor doubling time is 20.3–103 months for benign and 5.2–11.9 months for malignant lesions [36]. Given the small number of cases, the diagnosis of intraosseous leiomyoma requires a mitotic index no greater than 4 by 50 high power fields which is a criteria to differentiate soft tissue leiomyoma from leiomyosarcoma [19].

Leiomyoma being a benign tumor requires surgical excision either through intraoral or extraoral approach with negligible recurrence that is usually confined to vascular variants [37].

Conclusion

Intraosseous leiomyoma, though an uncommon tumor, is known to occur in the jaws. It shows a preference for the mandibular posterior region and a predilection for males. Hence it should be considered as one of the differential diagnosis of the jaw swellings. The treatment of choice is surgical excision with a very low recurrence rate.

Compliance with Ethical Standards

Conflict of interest None.

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Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed Consent Informed consent was obtained from the patient.

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