ISSN- O: 2458 - 868X, ISSN–P: 2458 – 8687
National Library of Medicine - ID: 101731606

International Journal of Medical Science and Innovative Research (IJMSIR)
IJMSIR : A Medical Publication Hub
Available Online at: www.ijmsir.com
Volume – 7, Issue – 2, March – 2022 , Page No. : 245 - 250

Odontogenic myxoma – case report and radiographic insight

[1]Kannampurath Arathi, Junior Resident, Department of Oral Medicine and Radiology, Government Dental College, Gandhinagar, Kottayam 686008, Kerala, India.

[2]Ravi Aparna, Junior Resident, Department of Oral Medicine and Radiology, Government Dental College, Gandhinagar, Kottayam 686008, Kerala, India.

[3]Leela Srikantannair Sreela, MDS, Ph. D, Professor & Head of the Department, Department of Oral Medicine and Radiology, Government Dental College, Gandhinagar, Kottayam 686008, Kerala, India.

[4]Sivaprasad Twinkle, MDS, Additional Professor, Department of Oral Medicine and Radiology, Government Dental College, Gandhinagar, Kottayam 686008, Kerala, India.

Corresponding Author: Kannampurath Arathi, Junior Resident, Department of Oral Medicine and Radiology, Government Dental College, Gandhinagar, Kottayam 686008, Kerala, India.

Citation this Article: Kannampurath Arathi, Ravi Aparna, Leela Srikantannair Sreela, Sivaprasad Twinkle, " Odontogenic myxoma – case report and radiographic insight", IJMSIR- March - 2022, Vol – 7, Issue - 2, P. No. 245 – 250.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Odontogenic myxomas often have a distinctive radiographic presentation described as a "tennis racket", "honeycomb" or "soap bubble", pattern. Less frequently, examples of odontogenic myxomas with a "sunray" or "sunburst" pattern have been reported. Because malignant entities such as osteosarcomas more classically present with a sunray/sunburst appearance, odontogenic myxomas are rarely considered in the radiographic differential diagnosis of a sunburst lesion. The objective of this paper is to report a case of an odontogenic myxoma presenting with the sunburst pattern in addition to the classic tennis racket trabeculae.

Keywords: Benign, Myxoma, Odontogenic tumor, Sunburst.

Introduction

Odontogenic myxoma is an uncommon benign mesenchymal odontogenic tumor believed to arise from odontogenic ectomesenchyme. The evidence for its odontogenic origin arises from its almost exclusive location in the tooth—bearing areas of the jaws, its occasional association with missing or unerupted teeth and the presence of odontogenic epithelium. Clinically, odontogenic myxoma is a benign painless, locally invasive, slowly enlarging mass which accounts for 3-6% of all odontogenic neoplasms but shows wide variability in the clinical and radiological presentation. Radiographically, Odontogenic Myxoma can appear as unilocular or multilocular radiolucencies, and has well-known patterns including "soap bubble", "tennis racket", and "honeycomb"[1].

The "sunray" or "sunburst" radiographic appearance of OMs has been infrequently reported in the literature.

Histopathologic ally, odontogenic myxoma is characterized by loose, abundant mucoid stroma that contains rounded, spindle shaped or angular cells.

Cellular and nuclear pleomorphism is rare, as is mitotic activity. The stroma may be relatively avascular or may exhibit delicate capillaries. Various surgical procedures were mentioned which includes curettage, excision (0.5 mm from apparent normal bony margin), and resection (1 cm from apparent bony margin)[2]. Radical treatment of block resection is advised by most authors over conservative treatment due to its invasive nature, large size, and recurrence history even though this intervention poses patients post treatment rehabilitation difficulties. The conservative management of myxoma by excision and curettage with liquid nitrogen cryotherapy is an alternative method proposed to radical resection. Liquid nitrogen will eliminate any remaining neoplastic cells by bone devitalization without affecting the inorganic structure, thereby yielding new bone formation[3].

This case report illustrates an Odontogenic Myxoma displaying a radiographic sunburst appearance, and provide a review of the literature on the varied radiographic presentations, and discusses the prognosis and recurrence of this radiographic variant.

Case report

A 39-year-old male patient reported to the Oral Medicine and Radiology outpatient department complaining of a swelling over the right lower gum for the past 4 years. Initially the swelling was small in size and showed a gradual increase to its present dimensions. No history of pain/paraesthesia was reported. Patient's medical history was non-contributory.

Extraoral examination revealed a diffuse swelling over the right Para symphyseal region with normal overlying skin. Intraorally, a well-defined swelling measuring approximately 3.5 × 2 cm with normal overlying mucosa was noted from 42 to 45 region involving buccal gingiva and vestibule (Fig 1). It was non-tender and hard with focal areas of firmness. Nearly uniform buccal cortical expansion and mild lingual cortical expansion was noted. Clinically missing 43 and distal displacement of crown of 42 was present. A provisional diagnosis of odontogenic tumor/fibrosseous lesion was made.

Panoramic radiograph shows an ill-defined radiolucent lesion crossing midline extending from mesial aspect of 34 to mesial aspect of 45. Internal structure of the lesion shows multiple loculations towards the distal aspect at the level of 44 and 45. An ill-defined area of increased radiopacity noted within the lesion .43 appears to be radiographically missing (Fig 2).

Cone beam Computed tomography images of the mandible showed a multilocular lesion with a few straight internal septa resembling tennis racket appearance; expansion and destruction of buccal cortical plates was noted with extension of the septa beyond the peripheral margin of the lesion at right angles imparting hairbrush/sunburst appearance. 43 appears to be impacted within the lesion (Fig 3).

Radiographic differential diagnoses considered were odontogenic myxoma, ameloblastoma, ameloblastic fibroma, central giant cell granuloma and central hemangioma. Aspiration cytology (performed with 18 G needle) was negative. Hence a differential diagnosis of odontogenic myxoma and ameloblastic fibroma was made. An incisional biopsy was done. Histopathology specimen showed moderately collagenous connective tissue stroma interspersed with bony trabeculae. Amid the bony trabeculae, the connective tissue was delicately collagenous with moderate cellularity (stellate cells) giving a myxomatous appearance.

Within the connective tissue clustered odontogenic epithelial cells, osteoblast-lined bony trabeculae and osteolytic lacunae were seen. The final diagnosis was

odontogenic myxoma. Surgical resection of the lesion with a safety margin of 1.5 cm followed by reconstruction was suggested as the treatment.

Discussion

Odontogenic myxoma was termed initially as "myxofibroma" by Rudolf Virchow in 1863 owing to its histologic similarity to the mucinous substance present in the umbilical cord. It was renamed as "odontogenic myxoma" later in 1947 by Thoma and Goldman[2]. The reported prevalence is 0.04–3.7%, with a slightly higher frequency (0.5–17.7%) in Asia, Europe and America. The age group commonly affected is 22.7 to 36.9 years, with a distinct female predilection. It is rarely seen in patients younger than 10 years and older than 50 years of age. Mandible is frequently affected than maxilla and commonly involves premolar and molar areas[4].

Odontogenic myxoma generally presents as an asymmetry of the jaw from a painless, slow growing lesion with possible loosening or displacement of teeth and cortical expansion but rarely root resorption. It is occasionally associated with missing or unerupted teeth[3]. Ulceration of the overlying oral mucosa occurs only when it interferes with dental occlusion. Rapid growth and infiltration into neighbouring soft tissue may occur.

Reviews with significant number of cases are few and includes 60 cases by Mosquida-Taylor et al. (1997), 64 cases by Lu et al. (1998), 33 cases by Simon et al. (2004), 25 cases by Li et al., and 30 cases by Claudia et al. (2007). Most of these authors supported the occurrence in young adults and female predilection. Common anatomic site reported was mandible followed by maxilla except for Regezi et al. (1978) who documented an equal distribution. The characteristic features of the case reported here comply with the results registered in literature[5].

Two types of odontogenic myxoma are identified, tumors that arise exclusively in jawbones (the most common) and those derived from perioral soft tissues[6]. Histogenesis of the tumour, though a subject of debate, has two schools of thought: myxomatous change in odontogenic fibroma & an osteogenic origin for the extra gnathic type, though the mesenchymal portion of tooth germ, dental papilla and periodontal ligament are still favoured as the tissues of origin. Hyaluronic acid seems to have a definite role in the invasiveness of the tumour, with mutations of PRKAR type 1A protein kinase subunit reported to regulate tumorigenesis [7]. This tumor has an extremely rare malignant version called odontogenic myxosarcoma. Based on an extensive survey of these tumors, odontogenic myxomas were thought to invade locally with possible recurrence after conservative treatment but rarely, if ever, to cause distant metastases[8].

A study by Zhang et al. (2007) divides the various radiographic appearances of OMs into six types. The frst type (type I) is a unilocular radiolucency. Multilocular radiolucencies creating a "honeycomb", "soap bubble", or "tennis racket" appearance are categorized as type II. Type III includes lesions that involve local alveolar bone. Type IV lesions consist of those involving the maxillary sinus. Lesions that cause osteolytic destruction and "moth-eaten" margins are categorized as type V. Lastly, the sunray appearance is classifed as type VI[7]. Other authors use the terms "hairbrush" and "fishbone" in addition to "sunburst" when describing this radiographic pattern in their case of OM. "Sunray", "sunburst", and "hair-on-end" are all synonymous radiographic terms used to describe thin, straight spicules of bone caused by a periosteal bone reaction.

This spiculated pattern is most characteristic of very aggressive and fast-growing tumors including

osteosarcoma, chondrosarcoma, Ewing sarcoma, central hemangioma, and metastatic prostate and breast lesions. The peri coronal & mixed radiolucent-radiopaque types are rare[9].The classic tennis racket pattern with sun ray appearance noted in our case favoured the diagnosis of odontogenic myxoma.

According to Peltola et al; root resorption is rare. Odontogenic myxoma frequently scallops between the roots of adjacent teeth and tends to grow along the involved bone without same amount of expansion seen in other benign odontogenic tumors. They rarely cross the midline. Imaging using CT and MRI (high tissue signal on T2 weighted MR images is characteristic) aid in establishing the intraosseous extent of the tumor, cortical perforation, soft tissue involvement, and accurate resection margins[10].

An exact diagnosis is necessary to plan appropriate surgical and adjuvant therapy. Recommended therapy can vary from a conservative curettage (with or without cautery) to resection with generous amount of surrounding bone to ensure removal of myxomatous tumor that infiltrates the adjacent marrow spaces. Surgical specimen appears as a pale brownish gelatinous substance (tender coconut appearance), which is considered pathognomonic.

Recurrence rates ranging from 10 to 33% have been reported in cases treated with simple excision and curettage. Insidious local invasion into cancellous bone beyond the radiographically visible margins and absence of encapsulation are considered as reasons for the high recurrence [11]. MMP 9 induced high invasiveness of the tumor into neighboring tissues makes the surgical method a crucial determinant of recurrence.

Recurrence following incomplete removal would usually occur within 2 years but can occur much later [1]. Based on the limited number of cases, there does not appear to be an increased rate of recurrence associated with the sunray/sunburst radiographic appearance compared to more traditional radiographic presentations.

Conclusion

This rare benign odontogenic tumor shows widely variant radiographic patterns. The sunray or sunburst presentation of OMs is not the most common presentation. Very often, the sunray or sunburst appearance has been associated with a malignant process and prompts a differential diagnosis that gravitates toward malignant entities. Although in our patient the sunray appearance was noticed, the gross examination of the resection specimen suggested features of a benign myxoma. Correlation of clinical, radiographic and histopathological features is a requisite to affirm the diagnosis of odontogenic myxoma.

References

1. Speight PM, Takata T. New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours. Virchow's Arch. 2018;472(3):331.
2. Reddy SP, Naag A, Kashyap B. Odontogenic myxoma: Report of two cases. Natl J Maxillofac Surg. 2010 Jul;1(2):183–6.
3. Manne RK, Kumar V suneel, Venkata Sarath P, Anumula L, Mundlapudi S, Tanikonda R. Odontogenic Myxoma of the Mandible. Case Rep Dent. 2012 Jul 9;2012: e214704.
4. Chaudhary Z, Sharma P, Gupta S, Mohanty S, Naithani M, Jain A. Odontogenic myxoma: Report of three cases and retrospective review of literature in Indian population. Contemp Clin Dent. 2015 Dec;6(4):522–8.

5. Varun AI, Ramachandran S, Rajasekhar an A, Balan A. Odontogenic myxoma: An archetypal presentation of a rare entity. J Indian Acad Oral Med Radiol. 2016 Oct 1;28(4):465.
6. Chaudhary Z, Sharma P, Gupta S, Mohanty S, Naithani M, Jain A. Odontogenic myxoma: Report of three cases and retrospective review of literature in Indian population. Contemp Clin Dent. 2015 Dec;6(4):522.
7. Willems SM, Wiweger M, Roggen JFG van, Hogendoorn PCW. Running GAGs: myxoid matrix in tumor pathology revisited: What's in it for the pathologist? Virchows Arch. 2010;456(2):181.
8. Lemberg, m. a., Calonius, b. p. e., Mäkinen, j. e. a., Paavolainen, m. p. and Syrjänen, k. j. (1984), a case of malignant myxoma (myxosarcoma) of the maxilla. European journal of oral sciences, 92: 352-357.
9. Zhang J, Wang H, He X, Niu Y, Li X. Radiographic examination of 41 cases of odontogenic myxomas on the basis of conventional radiographs. Dento Maxillo Facial Radiol. 2007 Mar;36(3):160–7.
10. Kheir E, Stephen L, Nortje C, van Rensburg LJ, Titinchi F. The imaging characteristics of odontogenic myxoma and a comparison of three different imaging modalities. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013 Oct;116(4):492–502.
11. Chrcanovic BR, do Amaral MBF, Marigo H de A, Freire-Maia B. An expanded odontogenic myxoma in maxilla. Stomatologija. 2010;12(4):122–8.

Legend Figures

Figure 1: Intraoral photograph, showing buccal cortical plate expansion and clinically missing 43 and displaced 42.

Figure 2: Panoramic radiograph showing ill defined, multilocular radiolucent lesion in the body of mandible crossing the midline

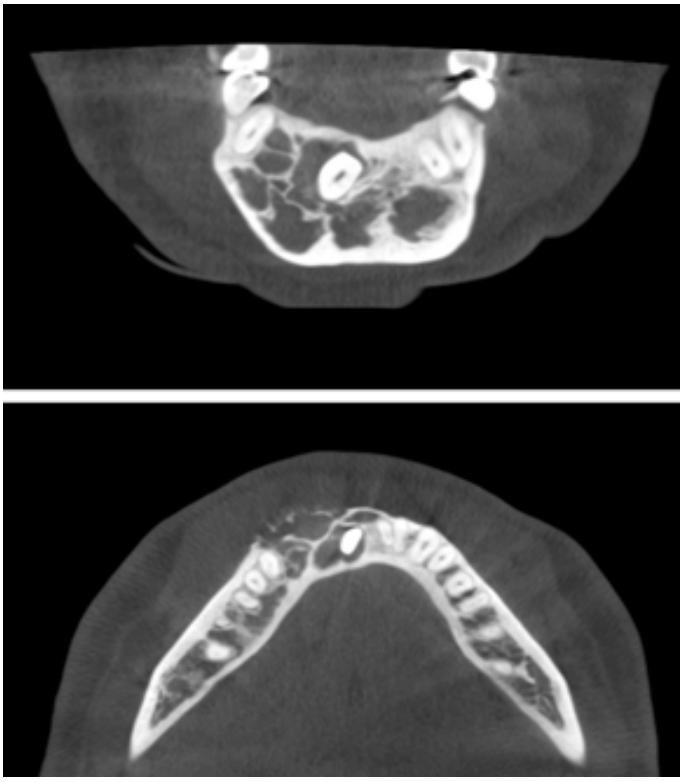

Figure 3 (a and b): CBCT images (Coronal and Axial section) showing tennis racket appearance with septa extending at right angles beyond the peripheral margin of the lesion imparting hairbrush/sunburst appearance

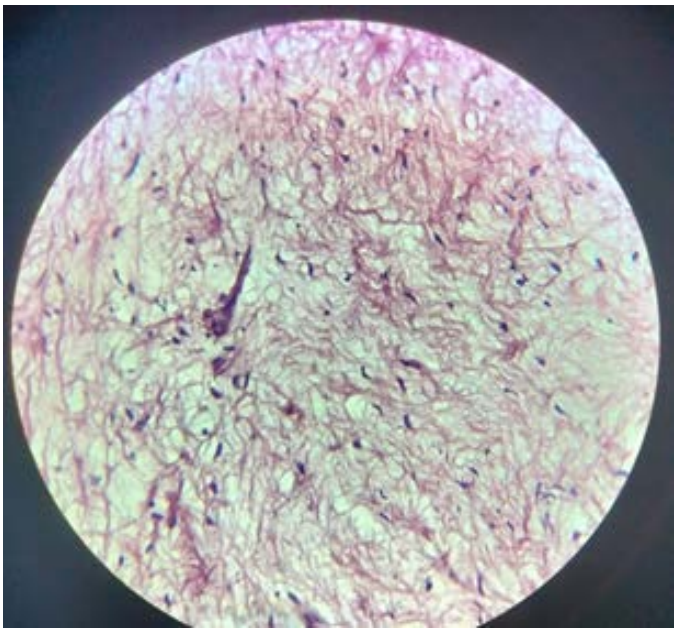

Figure 4: Histopathological view showing mixed area of fibrous tissue and inconspicuous stands of odontogenic epithelium in a myxoid stroma; magnification