Archives of Dental Research 2021;11(2):90–95

Content available at: https://www.ipinnovative.com/open-access-journals

Archives of Dental Research

Journal homepage: https://www.adr.org.in/

Review Article

A path less travelled — Short review on ossifying fibroma and osseous dysplasia

Deepali P Mohite[1],*, Devendra H Palve[1], Snehal R Udapure[1], Vinanti V Bodele [1], Mrunali D Jambhulkar[1], Vaishanavi A Borkar[1]

[1]*Dept. of Oral Pathology and Microbiology, Swargiya Dadasaheb Kalmegh Smruti Dental College and Hospital, Nagpur, Maharashtra, India*

ARTICLE INFO

Article history:
Received 25-07-2021
Accepted 10-08-2021
Available online 31-12-2021

ABSTRACT

Fibro-osseous lesions (FOL) are characterized by replacement of normal bone by fibrous tissue containing a newly formed mineralized product. The mineralized product may be ossification (sometimes cementum formation) or calcification of fibrous tissue by metaplasia. These lesions have similar radiographic and histopathologic features hence the term fibre- osseous lesion is not a diagnosis, rather a description of the presence of fibrous and calcified tissue. They may be developmental (fibrous dysplasia), reactive (cemento-osseous dysplasia) or neoplastic (ossifying/cementifying fibroma). This article presents the commonly used classification for fibro-osseous lesions and an insight into a few changes that have been introduced in the recent past with emphasis on Ossifying Fibroma and Osseous Dysplasia.

Ossifying fibromas are considered as benign fibro-osseous neoplasms which are principally encountered within the jawbones. Cemento-osseous dysplasias are non-neoplastic fibro- osseous lesion. Since 1971, the World Health Organization classified cemental lesions into 4 distinct entities, as follows: Periapical Cemental Dysplasia, Benign Cementoblastoma (true cementoma), Cementifying Fibroma and Gigantiform Cementoma (GC). The term, gigantiform cementoma, may imply a solitary process but it is misleading because the condition typically presents as slow-growing, multifocal/multiquadrant and expansile lesions involving both jaws. WHO classified it in Osseous dysplasia and El-Mofty et al. under Ossifying fibroma.

1. Introduction

Fibro-osseous lesions (FOL) are characterized by replacement of normal bone by fibrous tissue containing a newly formed mineralized product. Lesions may be developmental (hamartomatous), reactive, dysplastic or neoplastic. The ossification (sometimes cementum formation) or calcification of this fibrous tissue is due to by metaplasia in due course of time and most FOL's have similar radiographic and histopathologic features.[1] Montgomery is presumed to be the first to designate jaw lesions of central or intraosseous cemento-osseous lesions of jaws as ossifying fibromas.[2] Ossifying fibromas are considered as benign fibro- osseous neoplasms which are principally encountered within the jawbones.[3]

1.1. The various classifications of FOL proposed by different authors are listed below.[4]

1. 1985 — Charles Waldron[5]
2. 1987 — Working classification by Mico M. Malek
3. 1990 – Peiter J. Slootweg & Hellmuth Muller
4. 1992 — World health organization (WHO) classification
5. 1993 — Modified classification by Waldron

* *Corresponding author*.
E-mail address: deepali.mohite@sdk-dentalcollege.edu.in (D. P. Mohite).

https://doi.org/10.18231/j.adr.2021.014

6. 2001 — Brannon & Fowler classification[6]
7. 2005 — WHO classification of FOL[7]
8. 2006 — Paul M. Speight & Roman Carlos classification
9. 2008 - Eversole classification
10. 2017 – No specific modification

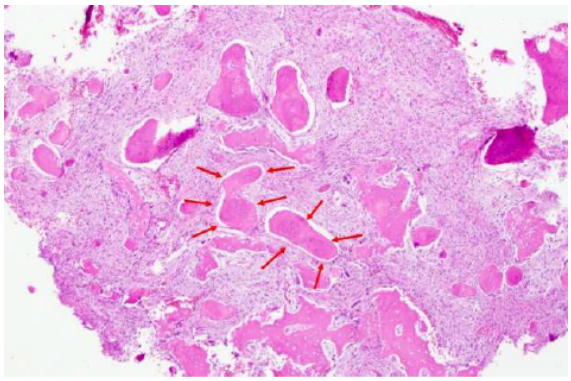

Fig. 1: Cemento-osseous dysplasia[4]

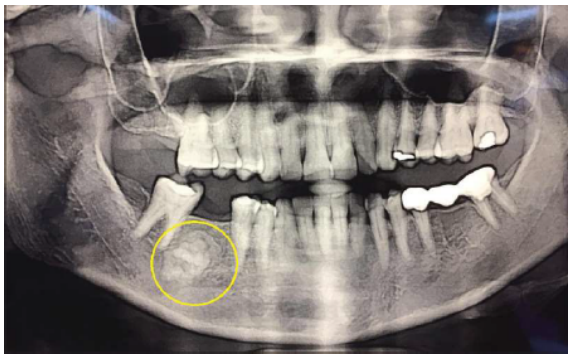

Fig. 2: Focal cemeno-osseous dysplasia — Orthopantamogram showing a well-defined radiopaque mass in the right mandible region extending from the distal root of 45 to the mesial root of 47.[16]

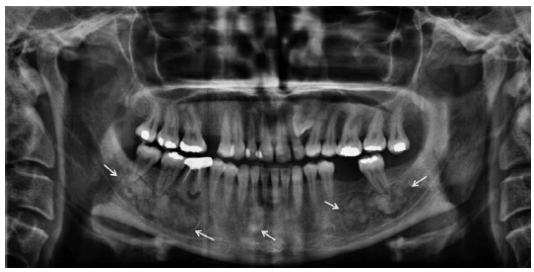

Fig. 3: Florid cemento-osseous dysplasia is situated at the right, left, and anterior portions of the mandible, surrounded by a radiolucent rim[9]

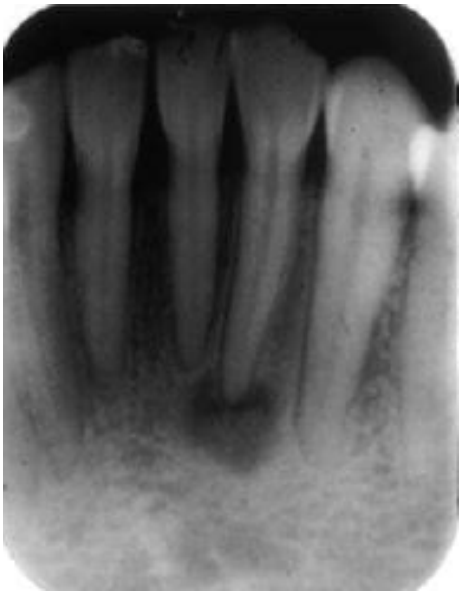

Fig. 4: Periapical cemento-osseous dysplasia — Periapical radiolucency[11]

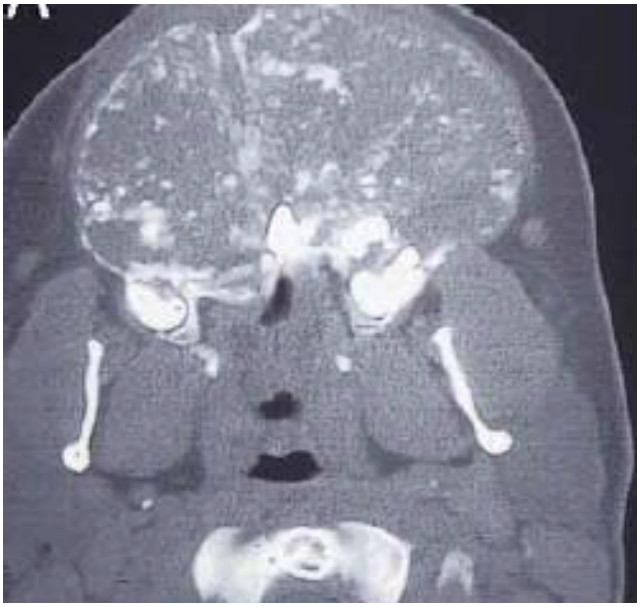

Fig. 5: Gigantiform cementoma : Axial CT images show extensive expansile mixed lesions of maxilla[12]

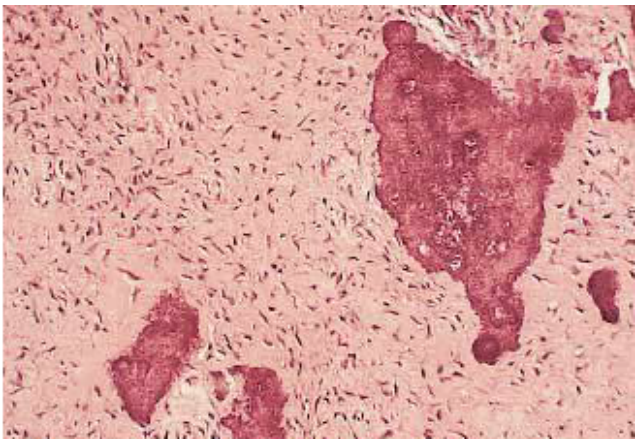

Fig. 6: Gigantiform cementoma: Proliferation of bland fibrous connective tissue with
spindle-shaped and stellate fibroblasts in hypovascular lesion with large cemental deposits.[12]

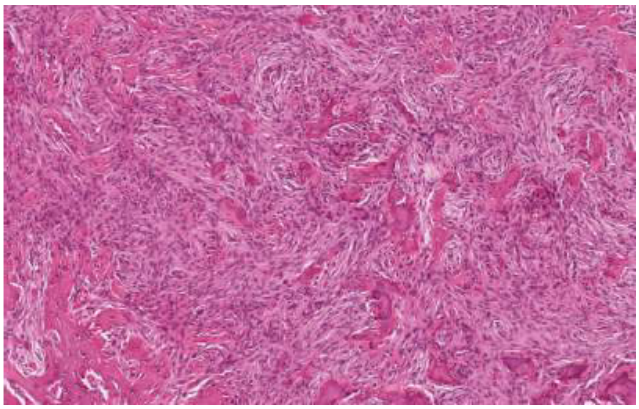

Fig. 7: Cemento-ossifying fibroma[13]

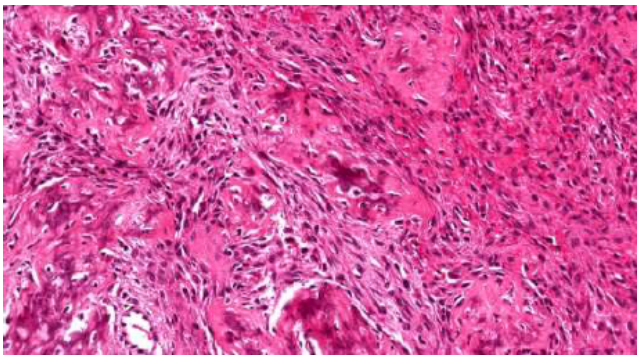

Fig. 8: Juvenile trabeculatr ossifying fibroma[13]

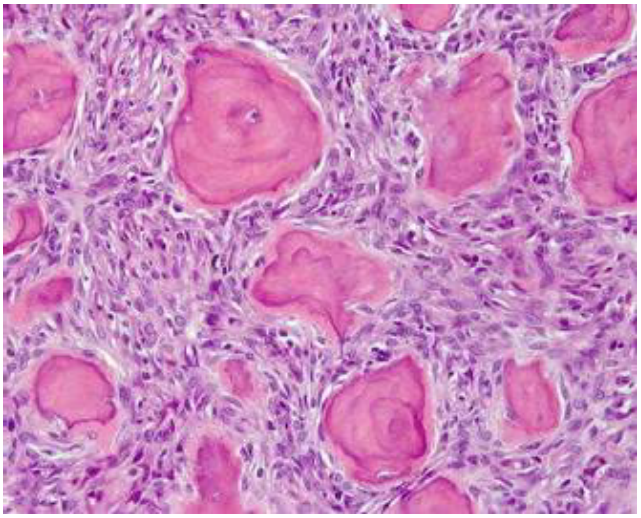

Fig. 9: Psammomatoid Juvenile Ossifying Fibroma[15]

1.2. Classification of fibro-osseous lesions — WHO classification of FOL (2005)[7]

1. Ossifying fibroma (OF)
2. Fibrous dysplasia
3. Osseous dysplasia
4. Periapical osseous dysplasia
5. Focal osseous dysplasia
6. Florid osseous dysplasia
7. Familial gigantiform cementoma
8. Central giant cell granuloma
9. Cherubism
10. Aneurysmal bone cyst
11. Solitary bone cyst

Osseous dysplasia also called as Cemento-Osseous dysplasia, Cemental dysplasia these are non-neoplastic fibro-osseous lesions of the tooth-bearing regions of the gnathic bones.[5] There is formation of cementum like mineralized component.2 (Figure 1)

1.3. There are 4 distinct forms

1. Focal cemento-osseous dysplasia
2. Florid cemento-osseous dysplasia
3. Periapical cemento-osseous dysplasia
4. Familial gigantiform cementoma

Focal cemento-osseous dysplasia is seen in middle-aged females commonly in mandibular molar region as an asymptomatic lesion small in size7 (Figure 2)

Florid cemento-osseous dysplasia was first reported by Melrose et al in 1976. It is asymptomatic and commonly presents as multifocal lesion in posterior maxilla and mandible region. Mostly often affects mandible (bilaterally) in middle aged or older black women. It presents as dull

pain and draining sinus associated with exposure of sclerotic bone.1 (Figure 3) Periapical cemento-osseous dysplasia (Periapical Cemental Dysplasia) It is present in apical region of vital mandibular anterior teeth as a solitary or multiple small lesions and exceed 1cm. It is asymptomatic, self-limiting, seldom lesion that expand the bone. (Figure 4) Familial gigantiform cementoma (Gigantiform cementoma) is a rare lesion first reported by Norberg in 1930. The term gigantiform cementoma, in as much as it implies a solitary process, is misleading because the condition typically resents as slow-growing, multifocal/multiquadrant and expansile lesions involving both jaws.12 It may arise from remnants of cementum which remain after extraction and is commonly seen in black females. It clinically presents as swelling of jawbones which lead to facial deformity, deposition of excessive cementum impairs vascularity of affected area, bone becomes susceptible to infection and complications like osteomyelitis may develop. It is inherited as an autosomal dominant trait. Lesions arise during childhood and progressively expand to cause facial deformity during early adult years.1,2,3

Radiographically appears as a radiolucent lesion in early stages, as the lesion starts to mature it shows radiopaque sclerotic border and mixed radiolucency - radiopacity. Fully mature lesions are radiopaque. It is restricted to the jaws and may arise in two, three or all four quadrants. The roots of the associated teeth are not affected and the lesion does not extend beyond the inferior alveolar canal or ramus though it crosses the midline of the jaws.1(Figure 5) Histopathological features show hypercellular fibroblastic stroma with monomorphic appearing spindle shaped fibroblasts and collagen fibers, immature bone trabeculae and cementum like tissue dispersed throughout the lesion. The latter is formed of hypocellular basophilic and curvilinear structures resembling cementicles that are normally seen in the periodontal ligament.1,3 (Figure 6)

The distinguishing features of types of osseous dysplasia are mentioned in Table 1 Cementifying fibroma, Ossifying fibroma, or Cemento-ossifying fibroma

It is fibro-osseous neoplasm, i.e. it is a benign bone-producing fibrous neoplasm of the skeleton. Ossifying fibroma is a well-demarcated lesion composed of fibrocellular tissue and mineralized material of varying appearances. [1]

Ossifying fibroma of the craniofacial skeleton are separated into two main clinicopathologic entities: [8]

Ossifying fibroma of Odontogenic origin (Cemento-ossifying fibroma), and Juvenile ossifying fibroma Cemento-ossifying fibroma (COF) synonymously called as Ossifying fibroma or Cementifying fibroma affects the tooth bearing areas of the mandible and maxilla. It is believed to be derived from the progenitor cells of the periodontal membrane which are capable of dual differentiation into fibroblasts, osteoblasts, and cementoblasts. Clinically it appears as a painless expansion of the jaws, particularly the mandible in individuals in the 3rd & 4th decades of life with a definite female predilection. [1]

The radiographic appearance shows well-defined and unilocular radiolucency which may be completely radiolucent or may show various degrees of opacification depending on the amount of calcified tissue present. In the mandible, larger lesions tend to expand inferiorly producing a characteristic downward bowing and thinning of the inferior border. Displacement of surrounding teeth and root resorption may be seen.2

Histopathologically the lesion is well-defined and may be encapsulated, showing hypercellular fibroblastic stroma with sparse collagen fibers and blood vessels, and containing variable amounts of calcified structures. Stromal fibroblastic cells show hyperchromatic nuclei. Calcified structures are composed of variable amounts of osteoid or bone and lobulated basophilic masses of cementum-like tissue resembling the cementicles that are normally found in the periodontal membrane. These structures may coalesce and form curvilinear trabeculae which may be acellular. Osteoblastic rimming of the bone trabeculae is evident. The cementum-like tissue is often woven and may show a characteristic quilted pattern.1,2 (Figure 7)

1. Juvenile ossifying fibroma (Table 2)
2. Its 2 forms are
3. Juvenile trabecular ossifying fibroma (JTOF)
4. Juvenile psammomatoid ossifying fibroma (JPOF)
5. Ossifying Fibromas Associated with Systemic Genetic Disorders

A group of benign fibro-osseous lesions affecting the jaws and microscopically described as ossifying fibromas or cemento-ossifying fibromas are manifestations of systemic genetic disorders. These are rare and not very well defined conditions.

1. Ossifying Fibromas Associated with Systemic Genetic Disorders are — 8
2. Hyperparathyroidism-jaw tumor syndrome
3. Familial gigantiform cementoma

Gnathodiaphysial dysplasia/fragile bone syndrome with fibro-osseous jaw lesions Hyperparathyroidism — Jaw Tumor Syndrome (HPT-JT) is an autosomal dominant syndrome with high but incomplete penetrance and variable expression, characterized by primary hyperparathyroidism (PHPT) with multiple parathyroid adenomas occurring at an early age with increased risk for parathyroid carcinoma, fibro-osseous jaw lesions and renal cysts or tumors. Jaw tumors occur during adolescence or early adulthood and they persist and may grow after parathyroidectomy and restoration of a normal serum parathyroid hormone level. Mutations in the HRPT2 gene have been reported to be associated with the occurrence of these lesions. The lesions

Table 1: Radiographical features of cemennto-osseous lesions

Focal cemento- osseous dysplasia	Periapical cemento- osseous dysplasia	Florid cemento- osseous dysplasia	Gigantiform cementoma
Single well defined lesion to teeth in posterior jaws	Well defined, Teeth of anterior mandible (vital)	Multiple sclerotic masses in tooth bearing areas surrounded by radiolucent zone, involves 2 or more quadrants of jaws	Multiple, irregular, oftenlobular radiopacities in maxilla and mandible, often crossses midline

Table 2: Features of Juveniletrabecular ossifying fibroma (JTOF) and Juvenile psammomatoid ossifying fibroma(JPOF)

Trabecular Juvenile Ossifying Fibroma	Psammomatoid Juvenile Ossifying Fibroma
Affects children and adolescents.	Affect 16 to 33 years.
Maxilla more frequent affected.	Particularly centered on periorbital frontal and ethmoid bones. Affects predominantly the extragnathic craniofacial bones.
Progressive & sometimes rapid expansion of the affected bone. In the maxilla, obstruction of the nasal passages & epistaxis may occur.	Bony expansion may involve the orbit or nasal bones and sinuses. Size from 2 to 8 cm in diameter. Expansion may result in proptosis, visual symptoms, and nasal obstruction.
Radiographic features are ground-glass and multilocular appearance. It is expansive and fairly well demarcated, with cortical thinning and possible perforation. Degrees of radiolucency or opacity depending on the amount of calcified tissue produced.	Radiographic features shows round well defined osteolytic lesion. Sclerotic changes may impart it with a ground glass appearance. May appear multiloculated on CT scans. Areas of low density due to cystic changes.
Histopathology - Loose architecture with hypercellular stroma composed of spindle cells with little collagen production. Osteoid develops directly from the fibrous stroma and forms long slender strands. Irregular mineralization takes place at the center of the strands resulting in the production of immature bone trabeculae that are devoid of osteoblastic rimming and do not show evidence of maturation. Aggregates of osteoclastic giant cells are typically found in the stroma. Occasional mitosis may be observed. Aneurysmal bone cyst formation has been reported in some cases. (Figure 8)	Histopathology – Unencapsulated & is significant for multiple small uniform ossicles (psammomatoid bodies) imbedded in cellular stroma composed of spindle & stellate shaped cells psammomatoid bodies are basophilic & bear some resemblance to dental cementum. At the periphery of the lesion these structures may coalesce and form bone trabeculae. Cystic degeneration and aneurysmal bone cyst (ABC) formation may occur.(Figure 9)
Treatment is excision.	Recurrence, even after definitive surgery. So no treatment.

are more common in mandible, occurring bilaterally and affecting the tooth bearing areas and may extend into the mandibular ramus. It is expansive, lytic lesions that may be unilocular or multilocular. It may be radiolucent, radio-opaque or a combination. Resorption of roots of the affected teeth is noted.

Histopathological features shows well-defined but unencapsulated and it is identical to COF. It composed of hypercellular fibroblastic stroma containing variable amounts of calcifying structures that may form woven bone trabeculae or cementum-like structures. Treatment is surgical.[8]

Familial Gigantiform Cementoma (FGC): FGC is a rare form of ossifying fibroma. Gigantiform cementoma is a rare, benign fibro-cemento-osseous disease of the jaws, seen most frequently in young girls. It is also included in this type of lesion.[5]

Gnathodiaphysial Dysplasia (GDD): It was first coined by Riminucci et al. Described as cemento-ossifying fibromas of the jaws associated with multiple fractures, bowing and cortical thickening of the tubular bone. It is a gigantiform cementomas associated with extragnathic skeletal abnormalities. Bone abnormalities are variably described as osteopenia, osteogenesis imperfecta, fragile and brittle bone. It is an autosomal dominant generalized skeletal syndrome. GDD1 gene, also known as TMEM16E or anoctamin 5 (ANO5) is mapped to a locus on chromosome 11p14.3–15.1. Two missense mutations (C356R and C356G) in GDD1. Radiographic features are expansive radiolucent with radiopaque areas. It affect both maxilla and mandible bilaterally. It shows multiple sclerotic areas of the tooth- bearing areas of the jaws or as a unilateral expansive multilocular lesion. The extragnathic bone shows osteopenia, demineralization, trabecular coarsening, diaphysial cortical thickening and bowing of the femora and tibiae. Histopathological features shows. Cemento- ossifying fibromas with immature bone and lobulated calcified masses in cell rich fibroblastic stroma. Treatment is surgical.[8–17]

2. Source of Funding

None.

3. Conflict of Interest

None.

References

1. Rajendran R. Shafer's textbook of oral pathology; 2009. p. 480–1.
2. Reichart PA, Philipsen HP, Allen CM, Neville BW, Damm DD. Odontogenic tumors and allied lesions.; 2004. p. 271–333.
3. Neville BW, Damm DD, Chi AC, Allen CM. Oral and Maxillofacial Pathology; 2002. p. 542–78.
4. Srichinthu K, Yoithapprabhunath R, Chitturi T, Yamunadevi RT, Potsangbam A, Singh AD, et al. Fibro Osseous Lesions-Classifications. *Int Biol Biomed J*. 2016;2(1):11–20.
5. Waldron CA. Fibro-osseous lesions of the jaws. *J Oral Maxillofac Surg*. 1993;51(8):828–63. doi:10.1016/s0278-2391(10)80097-7.
6. Brannon RB, Fowler CB. Benign Fibro-Osseous Lesions: A Review of Current Concepts. *Adv Anatomic Pathol*. 2001;8(3):126–69.
7. Thompson LD. World Health Organization classification of tumours: pathology and genetics of head and neck tumours; 2005. Available from: https://journals.sagepub.com/doi/pdf/10.1177/014556130608500201.
8. El-Mofty SK. Fibro-osseous lesions of the craniofacial skeleton: an update. *Head Neck Pathol*. 2014;8(4):432–76. doi:10.1007/s12105-014-0590-0.
9. Mortazavi H, Baharvand M, Rahmani S, Jafari S, Parvaei P. Radiolucent rim as a possible diagnostic aid for differentiating jaw lesions. *Imaging Sci Dent*. 2015;45(4):253–61. doi:10.5624/isd.2015.45.4.253.
10. Summerlin DJ, Tomich CE. Focal cemento-osseous dysplasia: a clinicopathologic study of 221 cases. *Oral surg oral Med oral Pathol*. 1994;78(5):611–31. doi:10.1016/0030-4220(94)90174-0.
11. Moft SKE, Bornstein MM, Reichart PA, Hürlimann S, Kuttenberger JJ. Periapical osseous dysplasia of the anterior maxilla–A case presentation; 2016. p. 1001–11.
12. Abdelsayed RA, Eversole LR, Singh BS, Scarbrough FE. Gigantiform cementoma: Clinicopathologic presentation of 3 cases. *Oral surg oral Med oral Pathol oral Radiol Endod*. 2001;91(4):438–44.
13. Gigantiform cementoma: clinicopathologic presentation of 3 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endodontol*. 2001;91(4):438–44. doi:10.1067/moe.2001.113108.
14. Martinez A. Juvenile ossifying fibroma. PathologyOutlines; 2021. Available from: https://www.pathologyoutlines.com/topic/mandiblemaxillajuvossifyingfibroma.html.Accessed.
15. Islam NM. Cemento-osseous dysplasia. PathologyOutlines; 2021. Available from: https://www.pathologyoutlines.com/topic/mandiblemaxillafloridcemento.html.Accessed.
16. Foss RD, Fielding CG. Juvenile Psammomatoid Ossifying Fibroma. *Head and Neck Pathol*. 2007;1(1):33–4. doi:10.1007/s12105-007-0001-x.
17. Ravikumar SS, Vasupradha G, Menaka TR, Sankar SP. Focal cemento-osseous dysplasia. *J Oral Maxillofac Pathol*. 2020;24:19–22.

Author biography

Deepali P Mohite, Reader

Devendra H Palve, Professor

Snehal R Udapure, MDS

Vinanti V Bodele , MDS

Mrunali D Jambhulkar, MDS

Vaishanavi A Borkar, MDS

Cite this article: Mohite DP, Palve DH, Udapure SR, VVB, Jambhulkar MD, Borkar VA. A path less travelled — Short review on ossifying fibroma and osseous dysplasia. *Arch Dent Res* 2021;11(2):90-95.