Case report

Clinico-Radio-Histopathological Aspects of a Compound Odontome with Impacted Deciduous Canine: An Uncommon Case Report

ABSTRACT

Odontomes are benign odontogenic tumors, mostly detected as incidental findings due to their slow growth and benign behaviour. They may cause complications such as delayed tooth eruption, impaction, deflection, or retention of teeth. Only 2% cases have shown impaction of a deciduous tooth. We hereby report a case of a compound odontoma in a 5-year-old child causing impaction of a deciduous canine.

Keywords: Compound Odontome, Primary dentition, Maxilla, Impacted deciduous

1. INTRODUCTION

The term “Odontome” was first coined by French physician Pierre Paul Broca in 1866 in his monograph ‘Traite Des Tumeurs’ to designate any tumour arising from odontogenic tissues. [1] However, with increasing cognizance, the usage of the term has gradually shifted and Odontomes are now considered as hamartomatous lesions of odontogenic origin, classified under ‘Benign Mixed Epithelial & Mesenchymal Odontogenic Tumors’ by WHO 2022 [2]; accounting for about 20% to 67% of all odontogenic neoplasms. [3,4]

Odontomas can occur at any age, but are typically identified within the first two decades of life [5]; with the mean age of detection being 14.8 years. [6] There is no gender predilection [3]. Normally, odontomas are small, but on rare occasions, they can enlarge and cause expansion of bones. [5] While the majority of odontomas are asymptomatic and are incidental findings on standard radiographs, a small number can even result in complications such as delayed tooth eruption, impaction, deflection, retention of deciduous teeth or infrabony defects. [7]Odontomes are common in the permanent dentition but rarely documented in conjunction with primary teeth. [8,9] The exact etiology of odontoma is not quite apparent [6] and has been associated with a number of local or genetic factors. [3]

The WHO categorizes odontomes into compound and complex types. [10] Compound odontomes are typically seen in the incisor cuspid region of the maxilla, whereas complex odontomes are more abundant in the molar and premolar regions of the mandible. [11] Compound odontomas demonstrate tooth-like features on radiography, encircled by a narrow radiolucent zone. Histopathologically, they consist of tiny "denticles" with observable morphologic differentiation and nearly normal dentin, cementum, and enamel matrix relationships. In contrast, complex odontomes appear as an irregular calcified mass surrounded by a thin radiolucent rim. Histopathologically, they are composed of a haphazard array of less differentiated odontogenic tissues, forming an amorphous mass. [4]

This report presents a rare case of compound odontoma associated with the primary dentition of a 5-year-old boy.

2. CASE REPORT:

Clinical findings:-

A 5-year-old male patient reported to the Department of Oral and Maxillofacial Pathology with the chief complaint of unerupted maxillary left deciduous canine and a gradually enlarging painless hard swelling in the region since the last 3 months. There was no history of trauma to the mouth or face and no significant medical history. Overall, the patient had good general health.

On extraoral examination, no obvious facial asymmetry was detected (Figure I [A]). Intraoral examination revealed the presence of unerupted left maxillary deciduous canine (63) and a fairly delineated, dome-shaped swelling in the left maxillary anterior region extending from the distal surface of left maxillary deciduous lateral incisor (62) to the mesial surface of left maxillary deciduous first molar (64), causing obliteration of labial vestibular sulcus. The overlying mucosa appeared smooth and stretched with normal physiological pigmentation (Figure I [B]). On palpation, the swelling was bony hard in consistency and no tenderness was elicited.

Diagnostic assessment:-

Panoramic radiograph revealed the presence of a well circumscribed, round to ovoid, radiopaque mass measuring about 2 cm x 1.5 cm, encased by a narrow radiolucent zone, in the maxillary left anterior region, obscuring the pathway of eruption of the unerupted primary canine (63). The radiopaque mass resembled a conglomeration of multiple small tooth-like structures (Figure II [A]).

Cone Beam Computed Tomography (CBCT) of maxilla revealed deciduous dentition in relation to 51, 52, 61, 62, 64, 65; crown formation in relation to 11, 12, 21, 22, 23, 24, 25, 26, 27 with incomplete root formation; vertical impaction in relation to 63; and an ill-defined, round to oval radiopacity with a well-corticated periphery measuring 23.7 mm (supero-inferiorly) x 17.6 mm (mesio-distally) x 15.5 mm (labio-palatally) noted at the level of crestal bone; extending from the distal aspect of 62 to the mesial aspect of 64, mesio-distally. The internal structure of the pathology was mixed radiopaque-radiolucent in nature, with the multiple small radiopacities resembling teeth. It was surrounded by a radiolucent area and an outer thin radiopaque rim (Figure III [B1-C2]). Expansion and thinning of the labial cortex with minute areas of breach were noted but the palatal cortex was intact. The highest position on the incisal tip of the crown of vertically impacted 63 having a single completely formed root was noted to be approximately 17.1 mm below the alveolar crest and abutting the labial cortex (Figure II [C3 & C4]). The palatal surface of the tooth was noted to be in approximation with left antrum and mesial surface abutted the lateral wall of left nasal cavity. The bone type was noted to be Lekholm& Zarb Type III bone.

Based on the clinical and radiographic findings, a provisional diagnosis of Compound Odontoma was made.

Surgical intervention & Histopathological evaluation:-

Surgical excision of the mass in toto was performed under general anaesthesia. Gross specimen resembled a conglomerated mass of malformed tooth-like structures (Figure III [A]).

The specimen was decalcified and processed using formalin fixation and paraffin embedding. Haematoxylin& Eosin-stained sections revealed the presence of tooth-like entities (Figure III [B]). They are composed of relatively homogenous dentinoid material showing dentinal tubule-like structures. Scant enameloid matrix was focally noted. They enclosed clefts containing pulp-like substance and was surrounded by a layer of fibrous connective tissue (Figure III [C-E]).

The overall histopathological features confirmed the diagnosis of “Compound Odontoma”.

Follow up &Outcome:-

Patient was advised periodic post-operative follow-up. Proper healing without any serious complications was noted. Patient was referred to Department of Pedodontia for further management and restoration of normal occlusal dynamics (Figure IV [A & B]).

3. DISCUSSION

Odontomas are mixed odontogenic tumors, that arise when fully differentiated epithelial and mesenchymal cells proliferate and develop into ameloblasts and odontoblasts. They are mostly composed of enamel and dentine, though variable amounts of pulp and cementum may be present [6,12,13] and are considered as hamartomatous malformations rather than true neoplasms. [14]

Various studies have attributed a number of causes to the etiopathogenesis of odontomas, including local trauma, infections and/or inflammatory conditions, remnants of cell rests of Serres, mature ameloblasts, odontoblastic hyperactivity, aberration in genetic control of odontogenesis or manifestations of hereditary anomalies like Gardner's syndrome, Hermann's syndrome among others. [3,4,11,15-18] Hitchin hypothesized that odontomas can arise from a mutation in a gene, a hereditary condition, or interference with the genetic regulation of tooth development, potentially occurring during the postnatal stage. [19] On the other hand, Levy has documented the traumatic injury-induced experimental induction of this lesion in rats. [20]

Xavier et al. demonstrated that a non-degradable variant of β-catenin expressed solely in SOX2-positive postnatal dental epithelial stem cells can activate WNT signalling aberrantly enough to develop malformations resembling odontomas. The mutant β-catenin in oral epithelium also induces ectopic expression of developmental signals like Shh and Bmp4 that impact adjacent tissues and promote abnormal mesenchymal proliferation and condensation. This significantly contributes to the formation of odontoblasts, dentine and pulp consistent with the tooth-like structures of odontomas. [21]

Odontomas can be diagnosed at any age, but are most frequently encountered in the first two decades of life. [5,7,22] According to studies by various authors, occurrence of odontomas have a male-to-female ratio ranging from 0.44:1 to 1.5:1 [4]; however, it is generally agreed that no significant gender bias exists. [3-5,16]

Compound odontomes are commonly located in the maxilla with a marked propensity for occurrence in the incisor-cuspid region. [5-7,23] They usually tend to remain asymptomatic, but may be associated with delayed eruption or impaction of teeth, and at later stages may occasionally effectuate enlargement of cortical bones and swelling. [3] Similar findings were reported in the case under discussion.Infection or regional lymphadenopathy may also be observed in severe cases.

Odontomes are rarely associated with the deciduous dentition. [8,9,15] In his review of 396 cases, Katz reported only 2% to be associated with the inability of a primary tooth to erupt. [24] The striking feature of the present case was its association with the primary dentition and the consequent impaction of a primary canine.

On radiographic analysis, compound odontomes reveal an irregular radiopaque structure with variable contour and size, composed of multiple tiny, malformed, primitive tooth-like entities, having a peripheral narrow radiolucent halo. [4,14,22]

Histopathological evaluation of compound odontomes show multiple structures having resemblance to miniature, single-rooted teeth comprised of a central pulpal core enclosed within a shell of dentin and partly covered by varying amounts of demineralized enamel matrix and cementum, contained within a capsule of loose fibrous connective tissue. [4,14,22] Identical radiographic and histopathological features were noted in the present case.

Since odontomes are benign encapsulated lesions, complete surgical enucleation is the preferred treatment. There is very little proclivity for recurrence. [11,22] Furthermore, it is crucial to conduct a thorough clinical and radiographic examination of the patient at regular follow-ups to evaluate any irregularity in the subsequent dentition.

4. CONCLUSION

Delay in eruption or exfoliation of primary teeth, or impaction of succedaneous permanent teeth should be meticulously evaluated clinically and radiographically to detect the presence of odontomes. Despite having limited growth potential, odontomes may lead to complications like cystic change or interference with normal occlusion which necessitates its surgical removal and subsequent periodic monitoring. Thus, regular assessment of dentition is of paramount importance as early diagnosis of odontomas permit simpler, less expensive treatment plans and ensures better prognosis.

CONSENT (WHEREEVER APPLICABLE)

All authors declare that 'written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal

ETHICAL APPROVAL (WHEREEVER APPLICABLE)

Ethical Committee Approval received

REFERENCES

1. Philipsen HP, Reichart PA. Classification of odontogenic tumours. A historical review. J Oral Pathol Med 2006;35(9):525-9.
2. Soluk-Tekkesin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2022 (5th) Edition. Turk PatolojiDerg 2022;38(2):168-184.
3. Ćabov T, Fuchs PN, Zulijani A, Ćabov Ercegović L, Marelić S. Odontomas: Pediatric Case Report and Review of The Literature. Acta Clin Croat 2021;60(1):146-152.
4. Boffano P, Zavattero E, Roccia F, Gallesio C. Complex and compound odontomas. J Craniofac Surg 2012;23(3):685-8.
5. Nelson BL, Thompson LD. Compound odontoma. Head Neck Pathol 2010;4(4):290-1.
6. Sivapathasundaram B, Preethi S, Rajendran R. Odontogenic Tumors. In: Sivapathasundaram B editors. Shafer's Textbook of Oral Pathology. 8th ed. India: Elsevier; 2016. p.117-8.
7. Isola G, Cicciù M, Fiorillo L, Matarese G. Association Between Odontoma and Impacted Teeth. J Craniofac Surg 2017;28(3):755-758.
8. Bordini J Jr, Contar CM, Sarot JR, Fernandes A, Machado MA. Multiple compound odontomas in the jaw: case report and analysis of the literature. J Oral Maxillofac Surg 2008;66(12):2617-20.
9. Kharbanda OP, Saimbi CS, Kharbanda R. Odontome--a case report. J Indian Dent Assoc 1986;58(7):269-71.
10. WHO Classification of Tumours Editorial Board. Head and neck tumours [Internet; beta version ahead of print]. Lyon (France): International Agency for Research on

Cancer; 2022. (WHO classification of tumours series, 5th ed.; vol. 9). Available from: https://tumourclassification.iarc.who.int/chapters/52.

11. Jain N, Pillai A, Varshini A, Kawadkar D, Dubey K, Singh A. Odontome- Revisted. J Clinical Research and Reports 2021;9(2).
12. Machado Cde V, Knop LA, da Rocha MC, Telles PD. Impacted permanent incisors associated with compound odontoma. BMJ Case Rep 2015 Jan 12;2015:bcr2014208201.
13. Das UM, Viswanath D, Azher U. A compound composite odontoma associated with unerupted permanent incisor: a case report. Int J Clin Pediatr Dent 2009;2(2):50-5.
14. Neville B.W., Damm D.D, Allen C.M, Chi A.C. Odontogenic Cysts and Tumors. In: Oral and Maxillofacial Pathology. 4th ed. Missouri: Elsevier; 2016. p.674-5.
15. Satish V, Prabhadevi MC, Sharma R. Odontome: A Brief Overview. Int J Clin Pediatr Dent 2011;4(3):177-85.
16. Rana V, Srivastava N, Kaushik N, Sharma V, Panthri P, Niranjan MM. Compound Odontome: A Case Report. Int J Clin Pediatr Dent 2019;12(1):64-67.
17. Sajesh S, Edsor E, Nandagopan S, Devi SS, Praveen V, Chidambareswaran S. Compound and Complex Odontomes: Case Series with Surgical Management and Brief Review of Literature. J Pharm Bioallied Sci 2021;13 Suppl 2:S1772-S1777.
18. Girish G, Bavle RM, Singh MK, Prasad SN. Compound composite odontoma. J Oral MaxillofacPathol2016;20:162.
19. Hitchin AD. The aetiology of the calcified composite odontomes. Br Dent J 1971;130(11):475-482.
20. Levy BA. Effects of experimental trauma on developing first molar teeth in rats. J Dent Res 1968;47(2):323-7.
21. Xavier G, Patist A, Healy C, Pagrut A, Carreno G, Sharpe PT et al. Activated WNT signaling in postnatal SOX2-positive dental stem cells can drive odontoma formation. Sci Rep 2015;5:14479
22. de Oliveira BH, Campos V, Marçal S. Compound odontoma--diagnosis and treatment: three case reports. Pediatr Dent 2001;23(2):151-7.
23. SolukTekkesin M, Pehlivan S, Olgac V, Aksakallı N, Alatli C. Clinical and histopathological investigation of odontomas: review of the literature and presentation of 160 cases. J Oral Maxillofac Surg 2012;70(6):1358-61.
24. Katz RW. An analysis of compound and complex odontomas. ASDC J Dent Child 1989;56(6):445-9.

FIGURE I: [A] EXTRAORAL PHOTOGRAPH; [B] INTRAORALLY, PRESENCE OF DOME-SHAPED SWELLING IN THE LEFT MAXILLARY ANTERIOR REGION

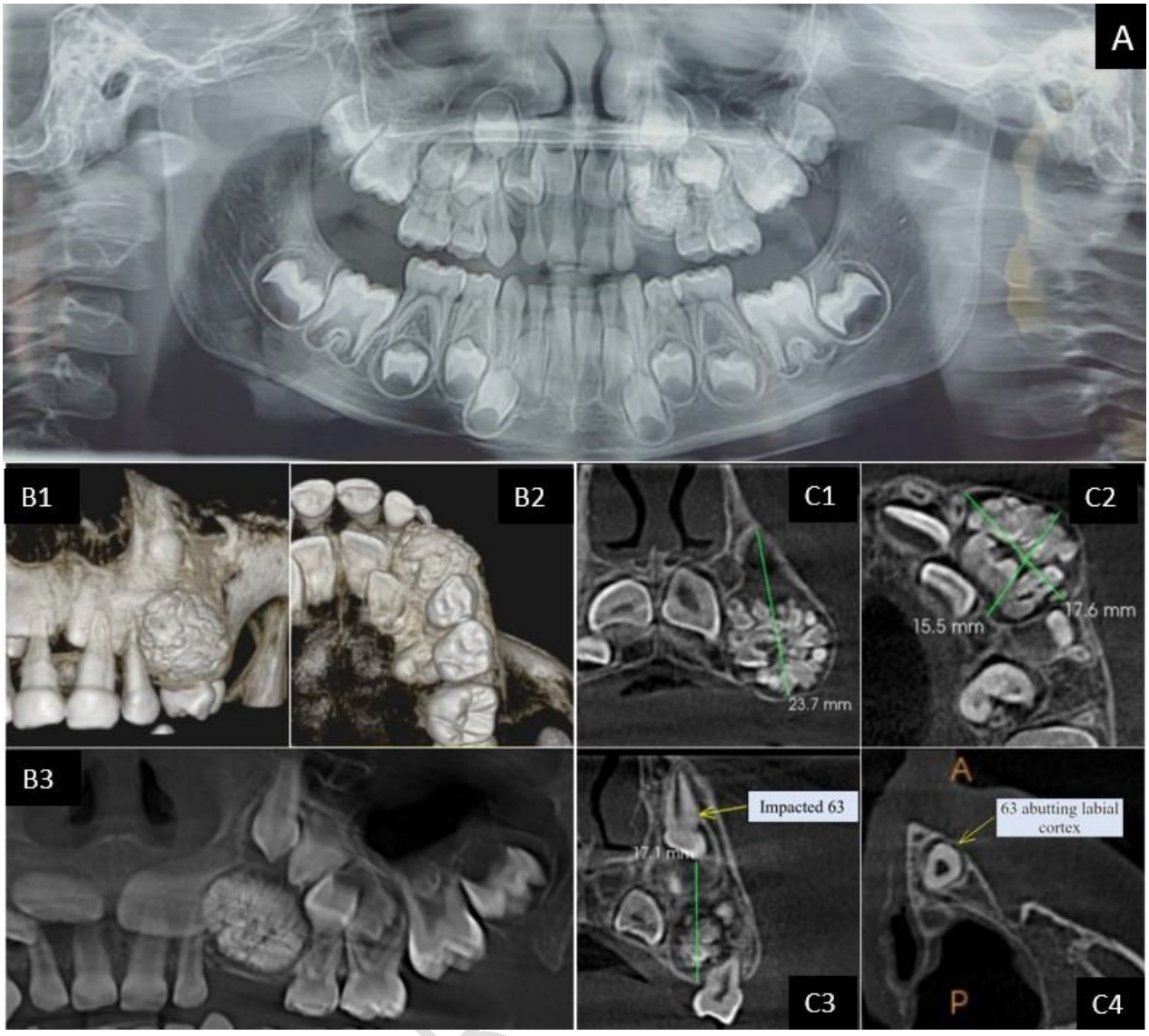

FIGURE II: [A] OPG SHOWING RADIOPAQUE MASS RESEMBLING MULTIPLE SMALL TOOTH-LIKE STRUCTURES IN THE MAXILLARY LEFT ANTERIOR REGION; RECONSTRUCTED CT IMAGES IN FRONTAL [B1] AND OCCLUSAL [B2] 3D VIEW AND PANORAMIC VIEW [B3]; [C1-C4] REFORMATTED MULTIPLANAR IMAGES THROUGH LEFT MAXILLA

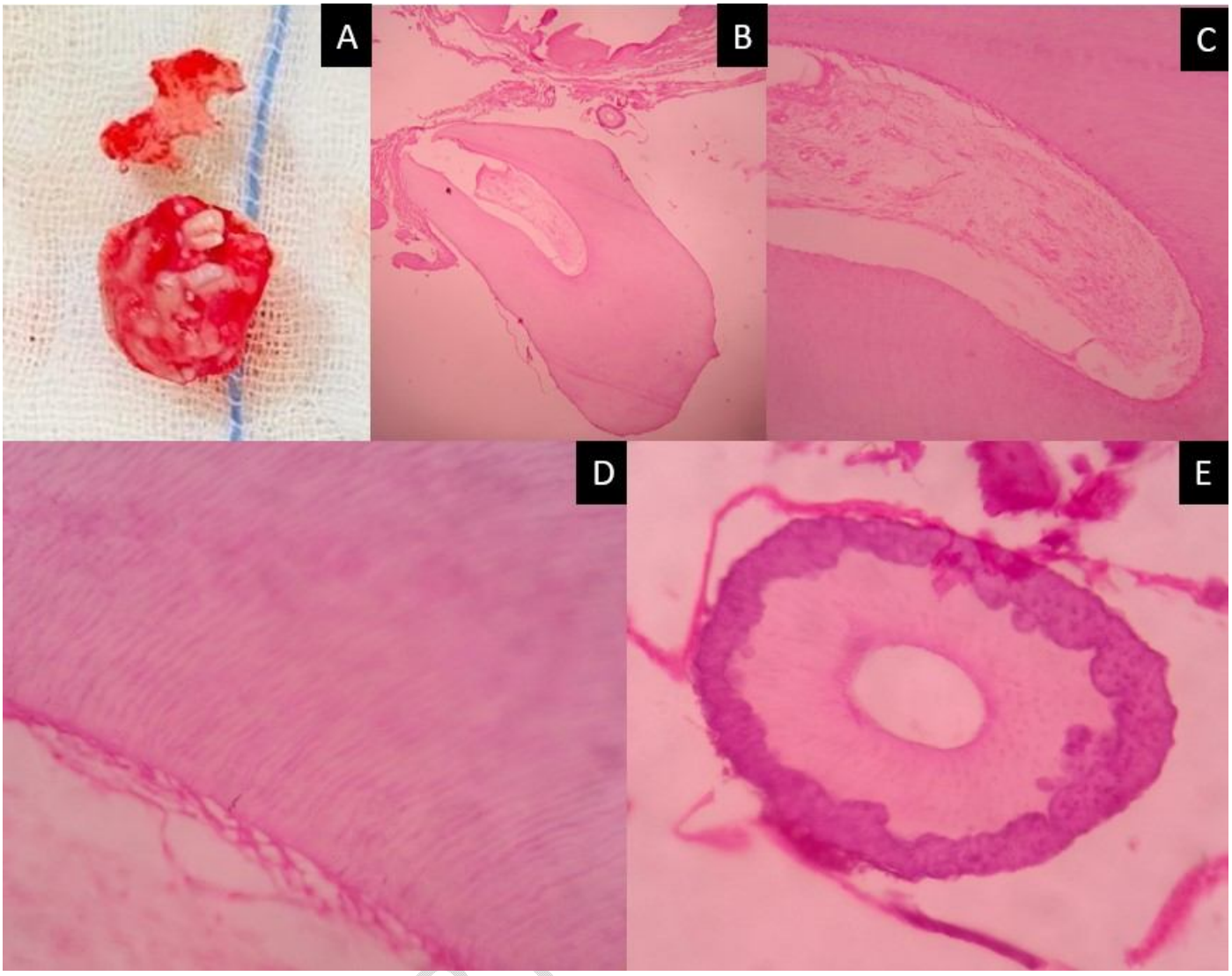

FIGURE III: [A] GROSS SPECIMEN; [B] 4X VIEW SHOWING TOOTH-LIKE ENTITY; [C] 10X VIEW SHOWING DENTINOID MATERIAL ENCLOSING PULP-LIKE TISSUE; [D] 40X VIEW SHOWING DENTINAL TUBULE LIKE STRUCTURES; [E] 40X VIEW SHOWING ENAMELOID AND DENTINOID MATRIX IN TRANSVERSE SECTION

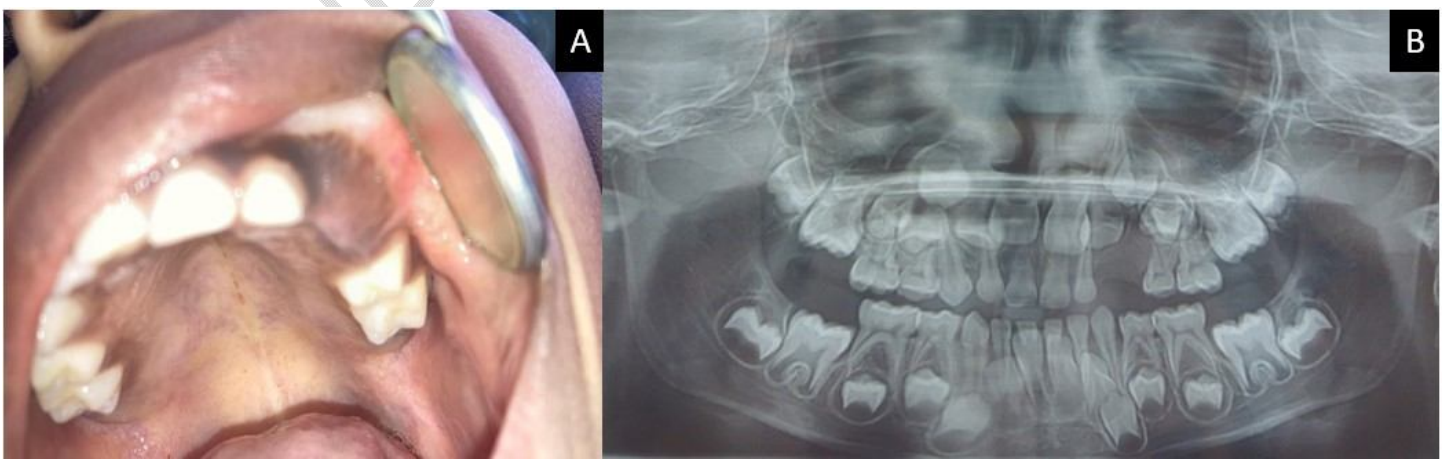

FIGURE IV: [A] POST-OPERATIVE PHOTOGRAPH; [B] POST-OPERATIVE OPG