CASE SERIES

Clinicopathological Correlation of Pericoronal Radiolucency in Pediatric Patients

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Abstract

An odontogenic epithelium-derived developing cyst of the mandible is known as an odontogenic keratocyst (OKC). According to the World Health Organization (WHO) classification from 2005, OKCs were referred to as keratocystic odontogenic tumors (KCOTs) and were thought to be cystic neoplasms. As of 2017, the WHO again switched KCOT back to OKC, as there was a lack of support for their justification as a tumor entity. This article presents a rare case report of single and multiple OKCs located in both the maxilla and mandible in male and female pediatric patients, demonstrating conservative as well as surgical management of OKCs. A dentigerous cyst was the provisional diagnosis in all three instances based on clinical and radiographic evidence. We planned both conservative and surgical treatment after considering the type and size of the lesion in the respective cases. A tissue specimen was then sent for histological examination, which confirmed our provisional diagnosis. Histological findings revealed a parakeratinized stratified epithelial lining with a thick columnar basal cell layer, surface corrugation, and neutrophils and lymphocytes organized against extravasated red blood cells (RBC), which confirmed the diagnosis of OKC in all three patients. Clinically and radiographically, OKC might resemble other benign, less aggressive tumors. Thus, it's critical to distinguish them from other cysts and tumors in order to provide appropriate care and lower the risk of recurrence.

Keywords: Case report, Jaw cyst, Multiple odontogenic keratocyst, Nonsyndromic, Pericoronal radiolucency.

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Introduction

Odontogenic keratocyst (OKC) arises from the remnants of the dental lamina and can occur singly or in large clusters of cysts, which are associated with Gorlin–Goltz syndrome. OKCs may be considered a benign neoplasm rather than a conventional cyst.[1] When it was originally identified by Philipsen in 1956, the pathology was classified as a cyst; however, this has since changed. In 2005, the World Health Organization (WHO) changed the classification of OKC to keratocystic odontogenic tumor (KCOT) due to the tumor's genetic behavior, aggressiveness, and infiltrative nature.[2] In 2017, the WHO once more reverted KCOT to OKC due to insufficient evidence supporting their classification as a tumor entity.[3]

Patients of all ages are affected by OKC, with the largest peak incidence (mean of 28.1 ± 18.3 years) occurring during the second and fourth decades of life. With a male to female ratio of 1.6:1, it is more prevalent among the white population. About 7.8% of all jaw cysts are OKCs, which also have a high recurrence rate (16.6–21.1%), especially in patients with numerous lesions, suggesting a higher risk of malignancy. In radiography, OKCs appear as unilocular or multilocular radiolucent images with distinct borders.[4]

Cysts can present in a variety of ways, so it's critical to provide an accurate diagnosis and appropriate treatment. The lesions clinically resemble one another, which presents a diagnostic challenge for clinicians. The most reliable method for identifying these lesions is still histopathology. Under the microscope, in addition to a palisaded base layer with basophilic nuclei, OKC has a characteristic lining of parakeratinized stratified squamous epithelium that may be aggressive and infiltrative.[5]

The literature describes a variety of OKC therapies, ranging from conservative methods to more intrusive operations. Conservative methods preserve tissue and reduce functional and esthetic harm.[6] Surgical intervention includes marsupialization (also called decompression), enucleation with or without adjunctive therapy (curettage, chemical cauterization, cryotherapy, electro cautery, and peripheral osteotomy), and resection. It has been proposed that using Carnoy's solution following surgical enucleation can stop recurrences. Because of the high recurrence rates of these lesions, a periodic follow-up is advised.[7]

The article presents a rare case series of three cases of OKCs in pediatric patients. The first case was of a single OKC located in the mandible, in a female patient. The second case was of multiple OKCs located in both the maxilla and mandible, in a male patient. The third case was in a female patient, consisting of multiple OKCs located in both the maxilla and mandible, presented to the Department of Pediatric and Preventive Dentistry, demonstrating conservative as well as surgical management of OKCs.

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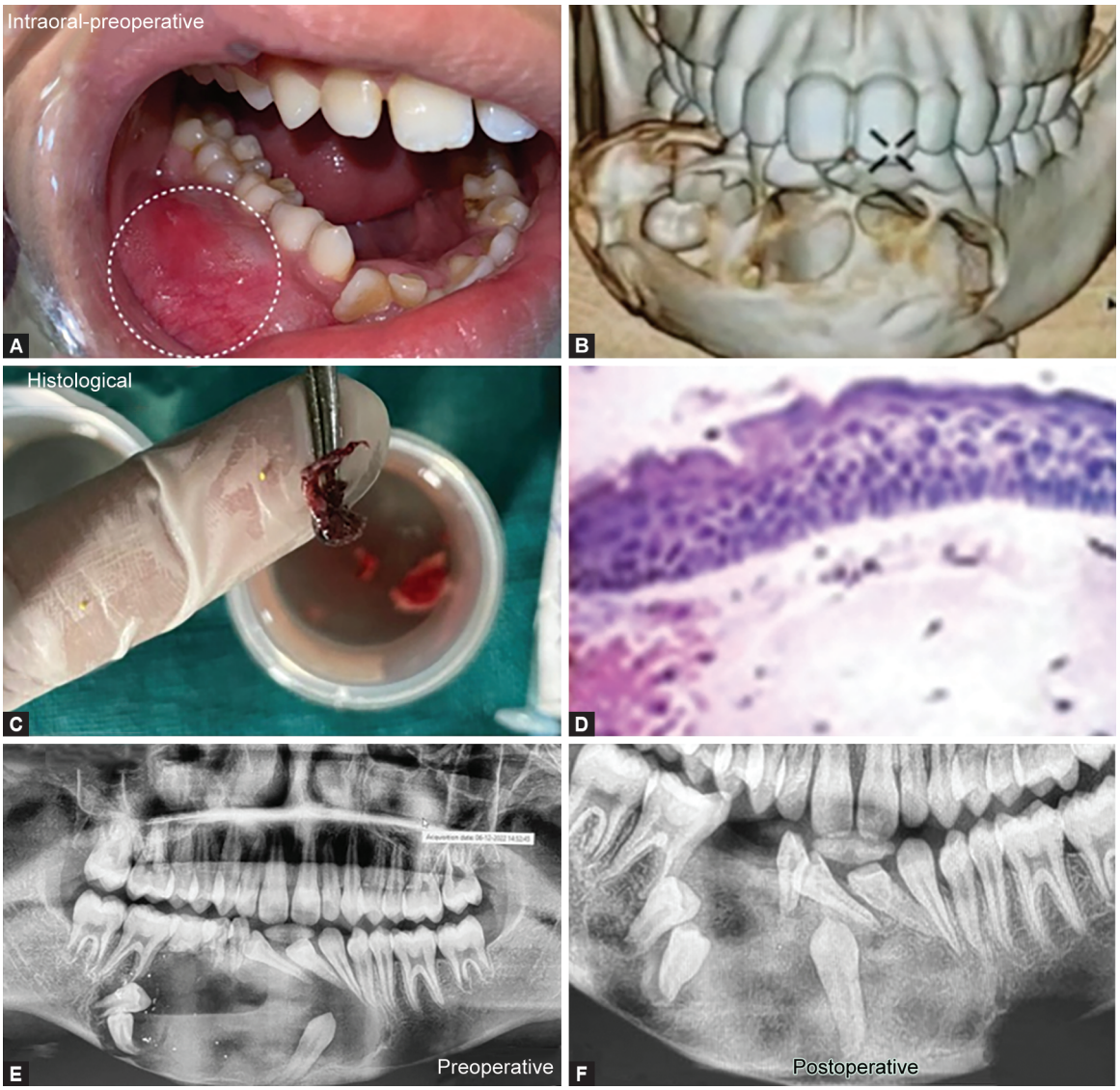
Case Series

Case 1

A 12-year-old female patient complained of a cystic lesion in her posterior right mandible for a year, which had radiological and clinical signs of a dentigerous cyst. On extraoral examination, there was facial asymmetry due to swelling detected on the right side of the mandible. Upon intraoral inspection (Fig. 1A), it was observed that the swelling extended from the right permanent lateral incisor to the right first molar, with the mucosa appearing to be in a healthy state. The swelling was painful, stable, and firm in consistency. The mandibular arch consisted of retained deciduous teeth: 71, 74, 81, 83, 84, and 85. On radiographic investigation, orthopantomogram (OPG) revealed a single well-defined pericoronal radiolucency in the right mandible measuring 3 × 3.5 cm, extending anteroposteriorly from the mesial aspect of 33 to the mesial side of 46. The pathology pushed the follicle of 43 and 44 downwards and backwards below the first permanent molar. The computed tomography (CT) report (Fig. 1B) revealed a huge expansile cystic lesion that covered the entire right half of the body, measuring around 3.3 × 5.5 cm. The buccal cortical plate was enlarged on the right side of the mandible. The provisional diagnosis of dentigerous cyst was made based on clinical and radiographic evidence.

After explaining the procedure to the patient's parents, a written informed consent was obtained. Because of the lesion's size, age, anatomical structures, and thin border of bone at the border of the mandible, which had a high risk of fracture on surgical intervention, an incisional biopsy followed by marsupialization was planned. Under local anesthesia, an incision was made to expose the lesion. The cyst was completely removed through a 2.5 × 1.5 cm window, and then Carnoy's solution was carefully applied. The lesion was then sent for histological analysis (Fig. 1C). The cavity was kept open using an iodoform dressing, which was replaced every 3rd day. A thermoplastic splint was delivered over the opening to prevent bacteria from entering the cavity. The biopsy finding (Fig. 1D) of parakeratinized stratified epithelial lining with a thick columnar basal cell layer, surface corrugation, neutrophils, and lymphocytes organized against extravasated red blood cells (RBC) confirmed the diagnosis of OKC in this patient. An OPG was performed after 4 months (Figs 1E and F), and the results showed

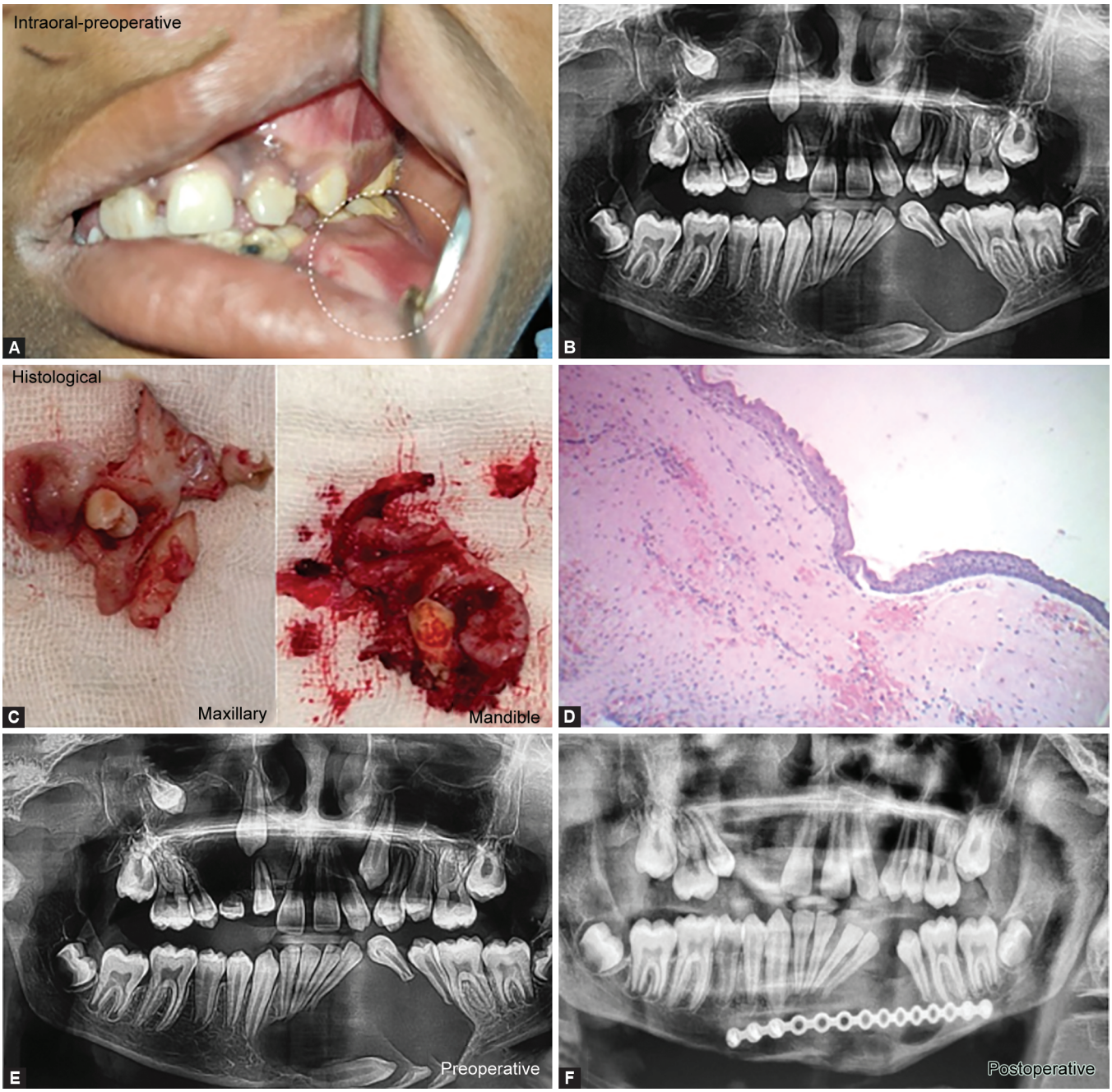


Figs 1A to F: Case 1: (A) Intraoral swelling; (B) CT revealed a huge expansile cystic lesion; (C) Removed lesion sent for histological analysis; (D) Histological findings; (E) Preop OPG; (F) Follow-up OPG after 4 months

increased bone density, a reduction in the size of the cystic lesion, and the impacted teeth's position shifted, indicating upright movement, with no evidence of recurrence and the patient being asymptomatic.

Case 2

A 13-year-old male patient presented with the main complaint of swelling on the lower left side of the mandible for a month. On extraoral inspection, diffuse swelling was noted on the left side of the lower border of the mandible, extending from the bottom region of the gingivolabial groove of teeth from the mesial aspect of 36 to 43. It was rigid, nontender, and nonfluctuant when palpated. On intraoral inspection (Fig. 2A), swelling was observed in the left mandibular region, and teeth from 36 to 43 did not respond to tests for pulp sensitivity, percussion, or palpation. Although the mandibular anterior teeth were crowded, there was no indication that the teeth were moving abnormally. On radiographic investigation, the OPG revealed (Fig. 2B) a large, distinct, and solitary radiolucent lesion in the mandible, involving teeth 36–43 (crossing the midline), and in the maxilla, involving teeth 12–15. The cone-beam computed tomography (CBCT) report revealed multiple cystic lesions: on the right side of the maxilla and maxillary sinus, a single well-defined pericoronal radiolucency measuring 45.6 × 26.2 mm, and the pathology had pushed the follicle of 13 and 14



Figs 2A to F: Case 2: (A) Intraoral swelling on left mandible region; (B) OPG revealed radiolucent lesions in the mandible, right, and left maxilla; (C) The maxillary and mandible specimen from the lesion was then sent for histological analysis; (D) Histological findings; (E) Preop OPG; (F) Follow-up OPG after 3 months

superiorly to the infraorbital region on the right side. On the left side of the maxilla, a single well-defined radiolucency measuring 14.6 × 13.7 mm was noted in the 23 region. On the anterior and left side body of the mandible, a single well-defined radiolucency measuring 53.9 × 28.0 mm extended from the 36 region to the 43 region (crossing the midline), with perforation and resorption of the buccal and lingual cortical plates. The pathology involved the left side mandibular canal. The provisional diagnosis of a dentigerous cyst was made based on clinical and radiographic evidence.

After explaining the procedure to the patient's parents, a written informed consent was obtained. Considering the multiple lesions and large size of the lesion, under general anesthesia, we intended to completely remove the lesion by enucleation and bone curettage. The surgical marking was done on the left side of the mandibular vestibule from 37 to 44 region (crossing the midline) and on the right and left sides of the maxilla from 16 to 12 region and 22 to 25 region, respectively. A full-thickness mucoperiosteal flap was raised in both maxillary and mandibular regions, and the lesion was enucleated along with affected teeth, that is, 12–14; 23, 33, and 34. The maxillary and mandibular specimens from the lesion were then sent for histological analysis (Fig. 2C). Chemical cauterization was done using Carnoy's solution, followed by copious irrigation using betadine solution in both maxillary and mandibular regions. A 15-hole plate of 1.5 mm was fixed using six (2 × 6 mm) screws in the left lower border of the mandible. The flap was sutured back with 3–0 Vicryl suture, and antibiotics and analgesics were recommended to the patient. Specimens from the lesions showed parakeratinized stratified epithelial lining with a thick columnar basal cell layer with hyperchromatic elongated nuclei. Loose to dense arrangement of collagen fibers with intense chronic inflammatory infiltrate, predominantly comprising of lymphocytes and zones of extravasated RBCs, confirmed the diagnosis of infected OKC (Fig. 2D). An OPG was performed after 3 months (Figs 2E and F), and the results showed increased bone density, upright movement of mandibular incisors, and also the typical eruptive force of 14 in the usual place, with no evidence of recurrence and the patient being asymptomatic.

Case 3

A 12-year-old female patient reported with the chief complaint of swelling on the left side of the infraorbital region. Extraoral examination showed swelling over the left maxillary area beneath the infraorbital region. It was firm, nontender, and nonfluctuant when palpated. On intraoral inspection (Fig. 3A), the mandible consisted of retained deciduous root stumps in relation to teeth 74, 75, 84, and 85, which demonstrated a negative response to percussion, palpation, and pulp sensitivity tests. On radiographic investigation, an OPG revealed a single well-defined pericoronal radiolucency in the maxilla involving teeth 24–26 with an unerupted tooth 25 and retained deciduous tooth 65. Additionally, a pericoronal radiolucency was also evident in the mandible involving teeth 31 to the mesial aspect of tooth 35. Retained deciduous root stumps were present in relation to teeth 74 and 75. The pathology pushed the follicle of teeth 34 and 35 downward and backward. An non-contrast computed tomography (NCCT) report (Fig. 3B) revealed a well-defined expansile cystic lesion measuring 3.0 × 2.3 × 2.1 cm in the alveolar process of the left maxilla, superiorly extending up to the left maxillary sinus and posteriorly up to the root of tooth 35. Another large expansile cystic lesion measuring 2.4 × 2.2 × 1.3 cm was seen in the left side of the body of the mandible, medially extending up to the left parasymphyseal region and posterolaterally extending up to the mesial aspect of the crown of tooth 35. The provisional diagnosis of a dentigerous cyst was made based on clinical and radiographic evidence.

After explaining the procedure to the patient's parents, a written informed consent was obtained. Marsupialization was planned because of the lesion's size, preference, and the thin border of the mandible. Extraction of retained deciduous teeth was done in relation to teeth 65, 74, and 75 under local anesthesia. In the maxilla, root canal therapy was attempted in relation to tooth 26, followed by Metapex dressing. The extraction socket of teeth 74 and 75 was kept open using an iodoform dressing, which was replaced every 3rd day, and the patient was instructed to maintain oral hygiene. The removed tissue was sent for histological analysis (Fig. 3C). At the subsequent visit, copious irrigation using betadine solution was performed in the mandibular region, and interrupted sutures were applied after inserting an iodoform gauze tail that made proximity with the socket wall. Histopathological findings (Fig. 3D) of the lesion revealed stratified epithelial lining with a thick squamous palisaded basal cell layer, surface corrugation, a loose to dense arrangement of collagen fibers with intense chronic inflammatory infiltrate predominantly comprising lymphocytes, confirming the diagnosis of OKC in this patient. In the maxilla, endodontic treatment was done in relation to tooth 26 (Figs 3E and F), but after that, the patient did not return because they belonged to another district. In a telephonic conversation, the patient's parent reported that there was no evidence of recurrence and the patient remained asymptomatic.

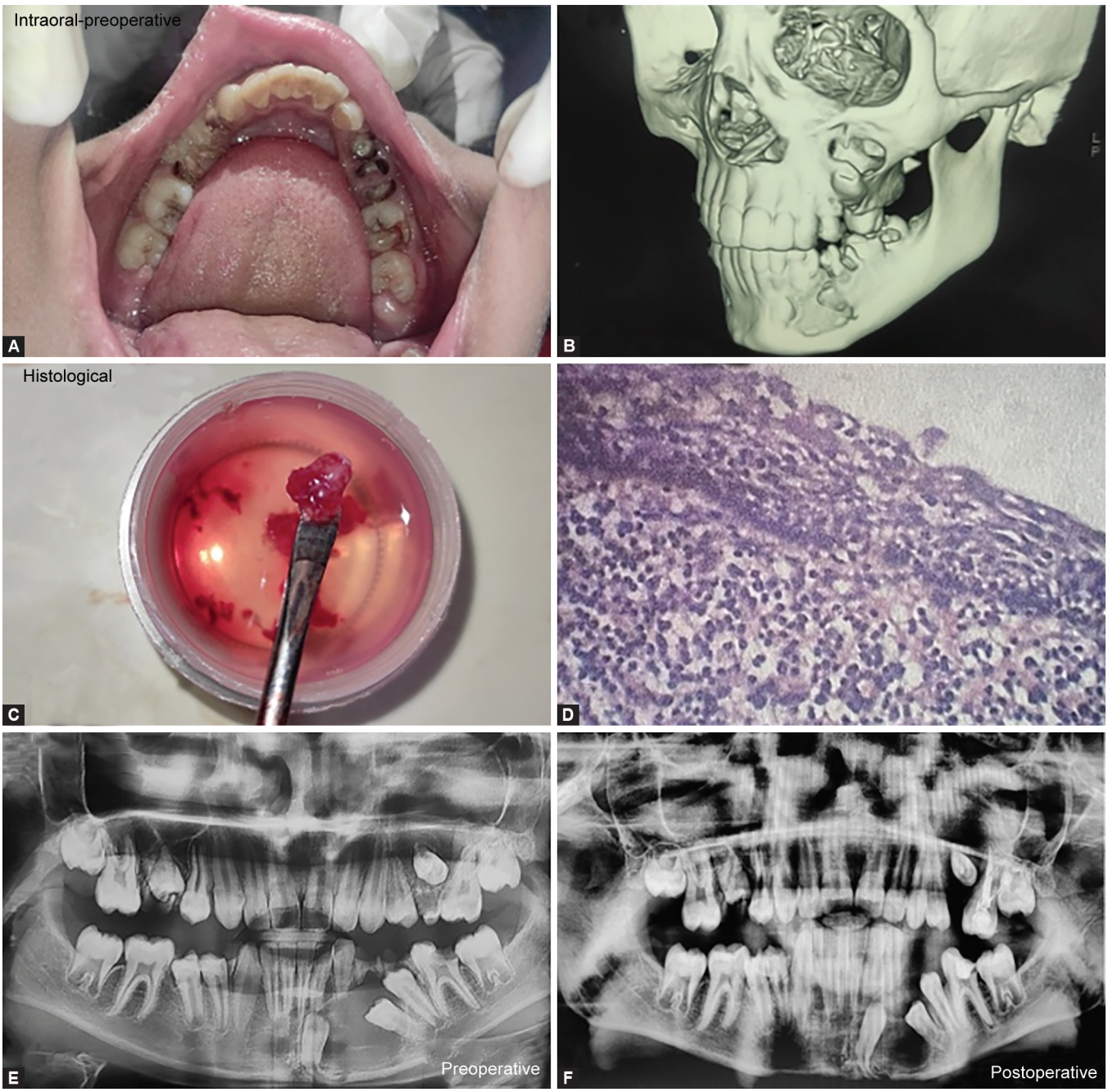
Discussion

Odontogenic keratocyst were reclassified as odontogenic developing cysts instead of benign odontogenic tumors due to new information about their morphogenesis and biological behavior. When OKCs are differentiated from other odontogenic cysts, such as dentigerous and radicular cysts, immunohistochemical expression of cytokeratin 17 and 19 is thought to be a better predictor. Additionally, their very aggressive behavior and recurrence are demonstrated by overexpression of proliferating cell nuclear antigen (PCNA) and Ki-67.[8]

Multiple OKCs usually occur as a component of nevoid basal cell carcinoma syndrome (NBCCS) or Gorlin–Goltz syndrome[9] and are also associated with other syndromes. Additionally, NBCCS may involve concurrent skeletal characteristics such as mandibular prognathism, frontal and parietal bossing, and bifid ribs, as well as cutaneous abnormalities like numerous basal cell carcinomas and palmar and plantar keratosis. There have also been reports of medulloblastoma, calcification of the falx cerebri, strabismus, mental impairment, and hypertelorism. None of these characteristics of NBCCS were present in our patient.

Brannon et al. found that, of the 312 OKC patients they examined, 5.1% had NBCCS and 5.8% had numerous keratocysts but no other syndrome symptoms.[10] The underlying genetic event is responsible for the molecular underpinning of the behavior linked to OKC. Both spontaneous and NBCCS-associated OKCs have a mutation in the PTCH gene 9q (22.3-q31), which is linked to NBCCS. The varied expression of NBCCS and sporadic OKCs has been attributed to a "two-hit" mechanism. Variability in the expression of the PTCH gene may be the cause of the lack of all NBCCS symptoms.[8]

The average age of presentation is in the third decade of life, although there is a large reported age range of 8–82 years. This



Figs 3A to F: Case 3: (A) Mandible consisted of retained deciduous root stumps irt 74, 75, 84, and 85; (B) NCCT revealed expansile cystic lesions in left maxillary and mandible region; (C) The removed tissue was sent for histological analysis; (d) Histological findings; (E) Preop OPG; (F) Postop OPG

report included situations involving adolescent groups. OKCs are intraosseous lesions that are mostly found in the posterior region of the jaws, primarily in the third molar, angle, and ramus in the mandible and the third molar/maxillary tuberosity in the maxilla. They are caused by the proliferation of remnants of the dental lamina or by the proliferation of cells from the basal layer of the oral epithelium into the mandible or maxilla. In contrast to the maxilla, the location is most frequently observed twice in the mandible.[11] As demonstrated by our patients, mandibular localization is more frequent than maxillary.

The following are examples of differential diagnosis: traumatic cyst (unilocular with scalloped margins, rarely shows cortical expansion), giant cell granuloma (usually in the anterior region of the jaw), ameloblastoma (usually multilocular, no straw-colored fluid on aspiration), dentigerous cyst (in OKC, the cyst is connected to the tooth at a point apical to the cementoenamel junction), and odontogenic myxoma. The most effective method for diagnosing an OKC is histopathological evaluation with an incisional biopsy. A folded, thin, regular parakeratinized epithelium that is 5–8 cell layers thick and devoid of rete ridges lines the fibrous cyst wall.[12]

The range of treatment options generally includes several invasive procedures like marginal or segmental resection as well as more conservative ones like marsupialization, enucleation,

curettage, chemical cauterization, and peripheral osteotomy. The lesion's location and size, its connections to neighboring structures, and whether OKC is primary or recurring all play a major role in the therapeutic decision. Because it preserves the growing dentition, minimizes cyst size, protects critical anatomical components (such as the sinus and inferior alveolar nerve), and promotes osteogenesis, marsupialization is characterized as a less aggressive method. A case of a large OKC treated by marsupialization and peripheral ostectomy was reported by Khalil et al.[13] in 2023. They concluded that a conservative and successful treatment for large OKC was marsupialization followed by peripheral ostectomy. On the contrary, enucleation is carried out when OKC is small and very slight harm to nearby tissues is anticipated. A mandibular OKC with aseptic pulp necrosis and substantial alveolar bone loss was reported by Tarallo et al.[14] in 2019. The OKC was entirely removed by enucleation and curettage, and the neighboring teeth were then treated endodontically. According to recent research, OKCs may be effectively treated by inhibiting the sonic hedgehog (SHH) signaling system.

Depending on the therapy approach, recurrence rates for OKC can range from 5% to over 70%. In their 2017 study, Chrcanovic and Gomez[15] examined the connection between surgical treatment and the recurrence rate of OKCs. They proposed that the recurrence of OKC could be caused by incomplete removal of the initial cyst, the growth of new OKC from satellite cysts or odontogenic epithelial remnants left in the surgical wound, or the development of a new lesion in nearby tissues that is thought to be a recurrence. According to Antonoglou et al.,[16] recurrences ranging from 17 to 56% occur after decompression, followed by enucleation or marsupialization. Although radical procedures like resections have a decreased chance of recurrence, they significantly impair a patient's appearance and functionality, which lowers their quality of life. However, after enucleation, adjuvant surgical techniques including curettage, chemical cauterization with Carnoy's solution, liquid nitrogen cryotherapy, or peripheral ostectomy are recommended to lower the chance of recurrence.

The professional must use sound judgment while assessing the lesion, its growth rate, and the extent of tissue invasion, as well as considerations such as patient cooperation and the requirement for long-term follow-up, in order to decide how best to treat a patient with pericoronal lesions.

Conclusion

Despite having common embryonic antecedents, pericoronal lesions exhibit a variety of variances. Clinicians may face a diagnostic conundrum when the radiological and clinical findings coincide. The therapy of such instances is based on an accurate diagnosis made through histological studies, followed by appropriate treatment and long-term follow-up.

Declaration of Patient's Consent

The authors attest to having acquired all necessary patient permission documents. The patient has consented on the form for the journal to publish their pictures and other clinical data. The patient is aware that while every attempt will be made to hide their identity, anonymity cannot be ensured and that their name and initials will not be published.

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