

## Case Report

# Large Keratocyst Extending to Mandibular Ramus and Coronoid Process: A Case Report

## *Mandibular Ramusa ve Koronoid Çıkıntıya Uzanan Geniş Odontojenik Keratokist: Olgu Raporu*

Büşra Şen<sup>1</sup> , Nesrin Dünder<sup>2</sup> , Elif Aslan<sup>3</sup> , Gözde Işık<sup>4</sup> , Dilara Özyiğit Büyüktalancı<sup>5</sup> 

### ABSTRACT

Odontogenic keratocysts are odontogenic epithelial-originated developmental cysts characterized by minimal expansion, high recurrence rate, and aggressive nature. Odontogenic keratocysts are usually observed as asymptomatic lesions detected during routine dental examinations; thus, they may be overlooked until the lesions reach large sizes. The treatment of odontogenic keratocysts is controversial, and surgical approaches range from conservative to more invasive procedures. In this case report, a 16-year-old female patient with an enlarged odontogenic keratocyst associated with an impacted third molar and extending through the mandibular angle, ramus, and coronoid process regions is presented with the clinical, radiological, and histopathological findings and the surgical treatment plan.

**Keywords:** Impacted third molar; Mandible; Odontogenic keratocyst

### ÖZET

Odontojenik keratokistler, odontojenik epitelden köken alan, minimal ekspansiyon ve yüksek nüks oranı ile karakterize gelişimsel kistlerdir. Odontojenik keratokistler, genellikle asemptomatik olmaları ve sıklıkla rutin diş muayenelerinde rastlantısal olarak tespit edilmeleri nedeniyle büyük boyutlara ulaşına kadar gözden kaçabilmektedir. Odontojenik keratokistlerin tedavisine dair farklı görüşler mevcut olup konservatif yaklaşımlardan daha invaziv prosedürlere kadar çeşitli cerrahi yaklaşımlar aracılığıyla tedavi gerçekleştirilmektedir. Bu olgu raporunda, 16 yaş kadın hastada saptanan, gömülü mandibular üçüncü molar diş ile birlikte izlenen ve mandibular ramus, angulus ve koronoid çıkıntı bölgelerine uzanım gösteren geniş bir odontojenik keratokist lezyonu klinik, radyolojik, histopatolojik bulguları ve cerrahi tedavi planı ile birlikte sunulmaktadır.

**Anahtar Kelimeler:** Gömülü üçüncü molar; Mandibula; Odontojenik keratokist

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İletişim: Dr. Büşra Şen

Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Ege University, Izmir, Turkey.

E-mail: [bsrasen93@gmail.com](mailto:bsrasen93@gmail.com)

<sup>1</sup> Araş. Gör. Dt. Ege University Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, Izmir, Turkey

<sup>2</sup> Prof. Dr. Ege University Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, Izmir, Turkey

<sup>3</sup> Uzm. Dt. Ege University Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, Izmir, Turkey

<sup>4</sup> Dr. Öğr. Üyesi Ege University Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Izmir, Turkey

<sup>5</sup> Uzm. Dr. Ege University Faculty of Medicine, Department of Pathology, Izmir, Turkey

## INTRODUCTION

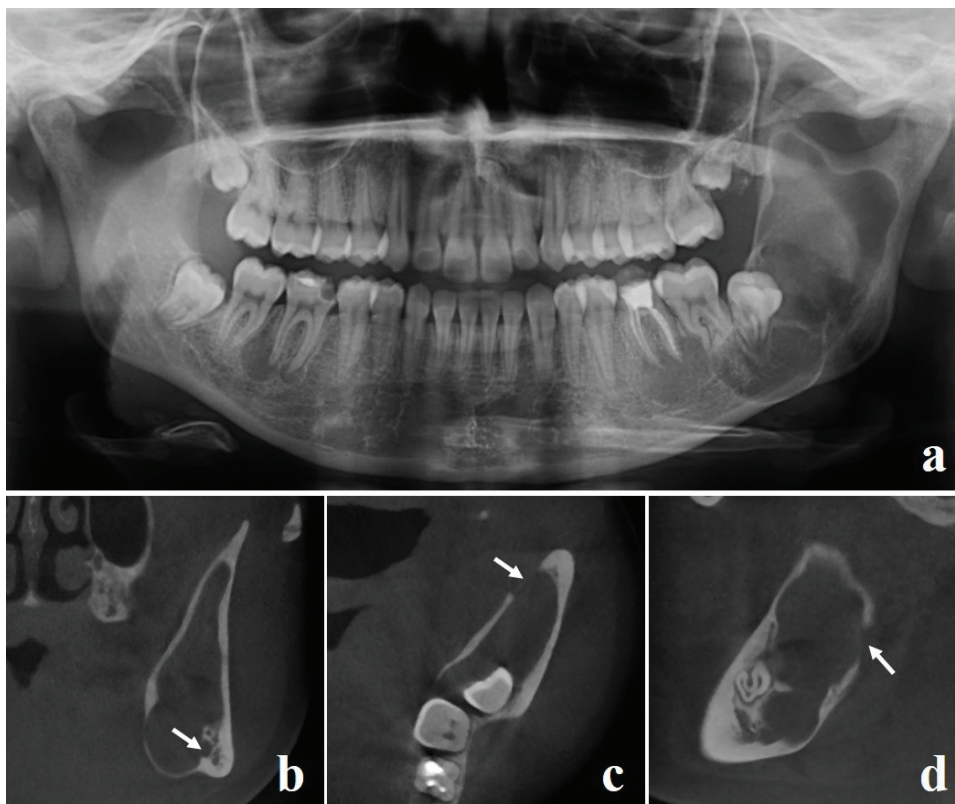
Odontogenic keratocysts (OKC) are developmental cystic lesions originating from odontogenic epithelial remnants in the alveolar tissue.<sup>1,2</sup> Unlike most odontogenic cysts, the enlargement of the keratocyst results from its innate growth potential rather than the internal osmotic pressure.<sup>2,3</sup> OKCs are often observed as asymptomatic and minimal expansive lesions located in the posterior region of the mandible and tend to expand towards the mandibular angle and ramus regions.<sup>1,3,4</sup> Some OKC lesions might be associated with an impacted tooth, and it may prevent tooth eruption in some cases.<sup>1-3,5</sup> Radiographic appearance of OKC has been defined as a well circumscribed, multilocular or unilocular, oval or round radiolucency.<sup>1-3</sup> Thin and keratinized structure of the epithelial tissue deriving from the dental lamina and the presence of satellite cysts are the distinguishing features of the keratocysts.<sup>2,3,5</sup> Nevertheless, histopathological examination is essential for the definitive diagnosis.<sup>2</sup>

In this case report, an overgrown OKC lesion associated with an impacted third molar and extending through the mandibular angle, ramus, and coronoid process regions is presented with the clinical, radiological, histopathological findings and the surgical treatment plan.

## Case Report

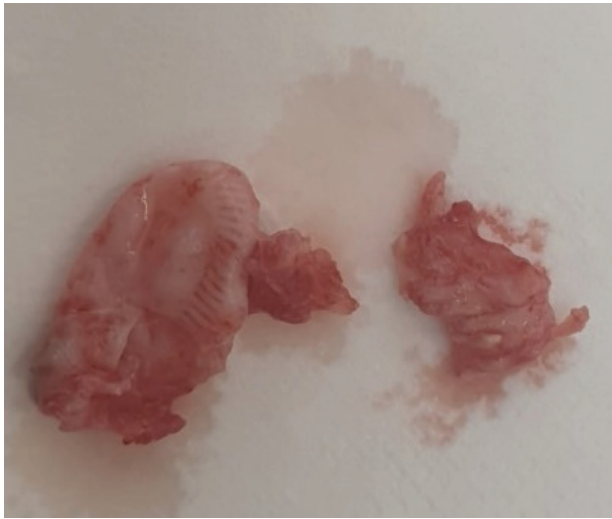
A 16-year-old female patient was referred to our out-patient clinic due to an intraosseous radiolucent lesion associated with the left mandibular impacted third molar detected during a routine dental examination.

The medical history did not include any systemic and/or syndromic disorders. Clinical examination did not reveal any intraoral and extraoral symptoms such as pain or palpable swelling. Radiographic examination with panoramic radiography and cone-beam computed tomography (CBCT) disclosed an intraosseous, multilocular radiolucent lesion associated with an impacted third molar in the left



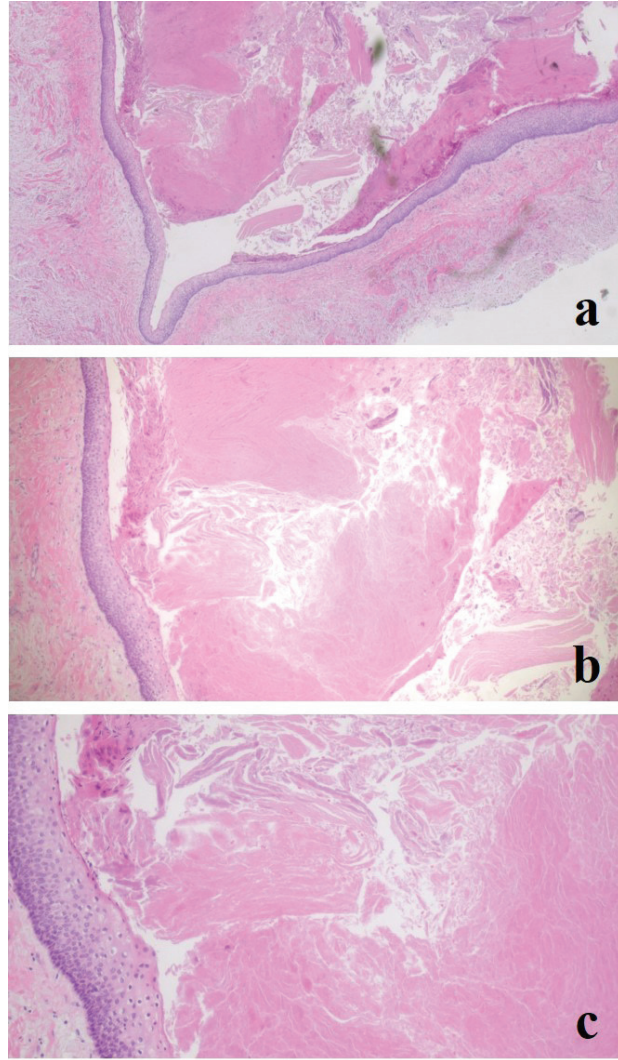
**Figure 1.** Preoperative panoramic radiography and CBCT sections. (a) Panoramic image showing mandibular ramus, angle, and coronoid process extension of OKC and the impacted mandibular left third molar, (b) Coronal CBCT section showing downward displacement of the mandibular canal, (c, d) Axial and sagittal CBCT sections showing lingual cortical bone destruction.

mandible. The lesion was extending through the left mandibular ramus, angle, and coronoid process regions (Fig. 1a). The dimensions of the lesion expanding along the anteroposterior direction of the mandible were measured as 19.23 mm x 48.75 mm, 10.73 mm x 35.17 mm, and 19.74 mm x 38.09 mm in the coronal, axial, and sagittal CBCT sections, respectively. The lingual cortical bone was destructed by the lesion in some regions and the mandibular canal was displaced downwards (Fig. 1b).



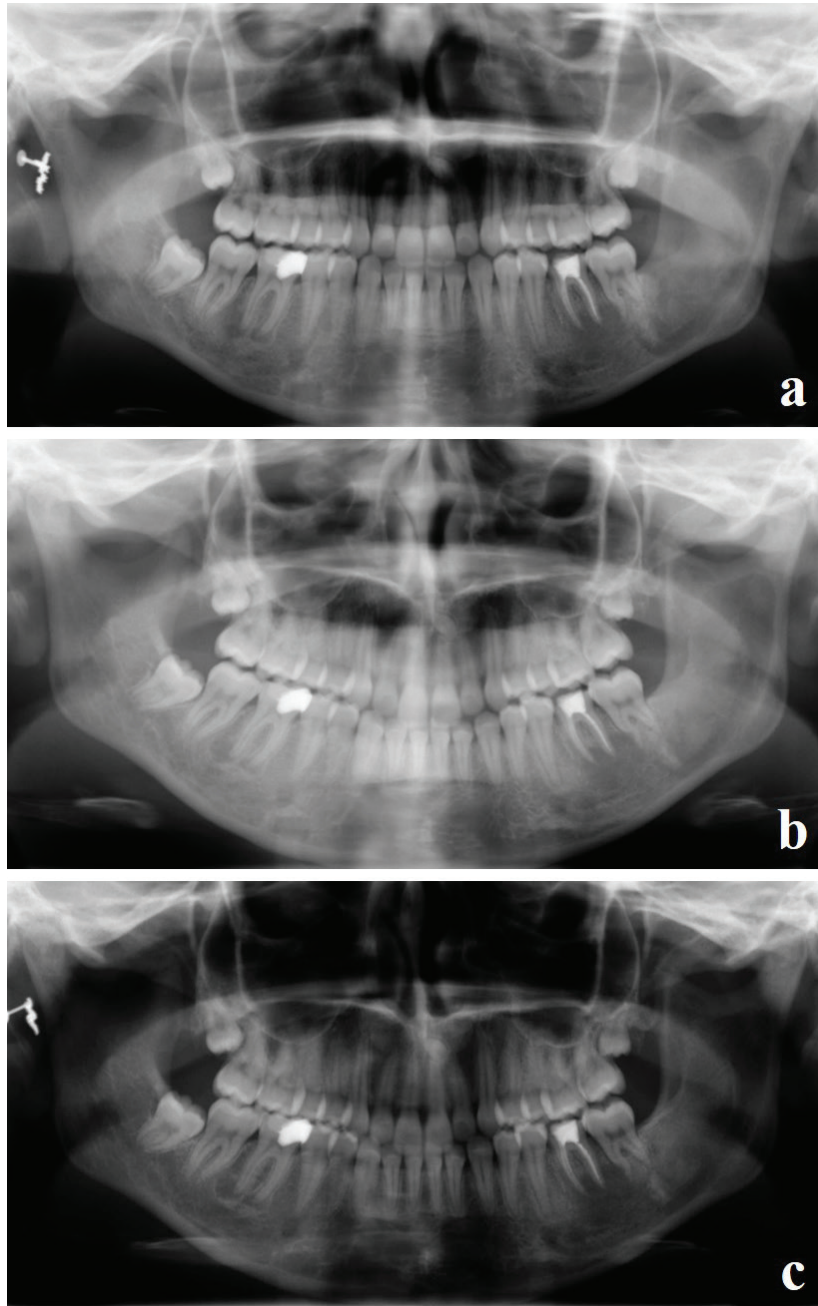
**Figure 2.** Tissue sample sent for histopathological examination.

The preliminary diagnosis was determined as OKC. Ameloblastoma and central giant cell granuloma were considered for differential diagnoses. The patient was referred to the Department of Oral and Maxillofacial Surgery for an initial biopsy and treatment planning. The initial biopsy was inconclusive due to insufficient biopsy material, and therefore, the total excision of the cyst epithelium was planned. Within the surgical treatment, extraction of impacted third molar, enucleation with curettage and peripheral osteotomy was performed under local anesthesia (lidocaine + 1/20.000 epinephrine), preserving vital anatomical structures. A horizontal incision was performed, starting from the ascending ramus, and continuing to the alveolar crest and the cervical lines of teeth #36 and #37. Then, the flap was elevated following a vertical incision performed in the mesial corner of tooth #36. Postoperative prescription included amoxicillin + clavulanic acid, metronidazole, naproxen sodium, and benzydamine. In addition, calcium and vitamin D complex was also prescribed to support bone healing.



**Figure 3.** Photomicrographs showing the histopathological findings. (a) Cystic structure containing keratinous debris in the lumen and surrounded by fibrous connective tissue around it (H&E, x40). (b-c) Keratocyst structure characterized by parakeratinized cyst epithelium lined with squamous epithelium and keratin lamellae in the middle (H&E, x100 and x200).

Epithelial biopsy material obtained from the mandibular ramus region was sent to the Department of Pathology in 10% formalin solution for histopathological evaluation (Fig. 2). The examination revealed a cyst lumen surrounded by fibrous connective tissue containing a keratinized debris and cyst epithelium lined with parakeratinized squamous epithelium characterized by keratin lamellae in the middle (Fig. 3). The final diagnosis was parakeratinized OKC. No recurrence was detected at the 3rd, 6th, and 12th month controls (Fig. 4). The patient was informed about the recurrence risk of the lesion and a 5-year long follow-up was planned.



**Figure 4.** Panoramic radiography images at the (a) 3rd month, (b) 6th month, (c) and 12th month postoperative controls showing the uneventful healing of the bony cavity.

## DISCUSSION

OKCs are odontogenic epithelial-originated cysts that can reach large sizes without any symptoms and tend to extend through the anteroposterior direction in the mandibular posterior regions. 25-40% of OKC lesions might be associated with an unerupted tooth.<sup>1-3,5,6</sup> It has been reported that OKC has a more rapid growing potential and tend to reach even larger sizes when associated with an impacted mandibular

third molar.<sup>7</sup> In agreement with this statement, we presented an enlarged OKC lesion associated with a mandibular third molar, extending from mandibular angle to coronoid process. It was noteworthy that OKC was incidentally detected during a routine dental examination, although it had grown to cover almost the entire ramus. Even though OKCs are generally localized in the mandibular posterior regions,<sup>1,5,8,9</sup> maxillary OKC lesions invading maxillary sinus have also been reported.<sup>10,11</sup> Therefore, aside from

the unerupted mandibular third molars, impacted maxillary third molars should also be considered for the risk of associated OKC lesions.

The treatment of OKC is controversial, and there are various surgical approaches recommended by different authorities.<sup>1,4,5</sup> Enislidis *et al.*<sup>12</sup> stated that there was no definitive method in the treatment of mandibular cysts reaching large sizes, and the size and histological structure of the cyst play a vital role in the decision of conservative or aggressive approach. The surgical treatment of OKC includes enucleation, marsupialization, decompression, resection, and reconstruction procedures combined with peripheral osteotomy, curettage, cryotherapy, carnoy solution, electrocautery, and 5-Fluorouracil.<sup>1-5</sup>

In the present case, enucleation under local anesthesia was preferred as the surgical treatment approach for the detected OKC lesion. In addition to enucleation, curettage and peripheral osteotomy were also applied in order to remove any epithelial tissue remnants and satellite cysts from the cavity, because the thin and fragile cyst wall of OKC increases the risk of recurrence.<sup>2</sup> Our treatment procedure was in agreement with the study of Al-Moraissi *et al.*<sup>4</sup> reporting that enucleation with peripheral osteotomy and curettage provides a 36.7% reduction in the recurrence rate compared to single enucleation of the lesion. When similar case reports presenting overgrown OKC lesions in the mandibular ramus and coronoid process regions were analyzed, it was noted that surgical resection and marginal mandibulectomy were the preferred treatment approaches in some of the cases<sup>6,8,13</sup> and under general anesthesia.<sup>9,14</sup> However, enucleation with peripheral osteotomy results in a successful healing process in OKC lesions,<sup>4,15,16</sup> and local anesthesia can be preferred to prevent the patients from taking unnecessary medication.<sup>17</sup>

The recurrence of OKC is usually observed within the first 5 years. However, the recurrence may also occur in a longer time following the surgery.<sup>2,3</sup> In the present case, an uneventful bone healing was noted, and no recurrence was detected at the 3rd, 6th, and 12th month follow-up. However, we are well aware that a long-term follow-up is required to confirm the success of the preferred surgical method, considering large cysts have a higher risk of recurrence.<sup>2,5</sup>

Therefore, a 5-year follow-up period was planned. Although recurrence has been generally considered as a failure of treatment, minor surgery and secondary curettage are shown to be effective in the management of recurrent OKC lesions.<sup>2</sup> Nevertheless, enucleation combined with peripheral osteotomy and curettage decreases the recurrence rate<sup>4</sup>

Differential diagnosis of OKC includes many intraosseous lesions such as dentigerous cyst, ameloblastoma, lateral periodontal cyst, odontogenic myxoma, simple bone cyst, radicular cyst, central giant cell granuloma, arteriovenous malformations and some fibroosseous lesions.<sup>1-3,5</sup> Rarely, epithelial tissue of keratocyst may show epithelial dysplasia and transform into squamous cell carcinoma.<sup>2,5,18</sup> In addition, it is also important to examine the patient for the presence of syndromes such as Gorlin Goltz Syndrome in the presence of multiple keratocysts.<sup>19,20</sup> In our case, the lesion had a radiolucent multilocular structure, minimal expansion, and was associated with an impacted tooth. Therefore, the preliminary diagnosis was determined as OKC, which was later confirmed by histopathological evaluation.

In conclusion, routine dental examination is critical regarding the early diagnosis of asymptomatic cystic jaw lesions such as OKC, which may be overlooked until it reaches very large sizes due to its asymptomatic and minimal expansive developmental nature. Therefore, in cases where an impacted third molar was detected, OKC should be considered as an underlying reason for the impaction.

## REFERENCES

1. Chirapathomsakul D, Sastravaha P, Jansisyant P. A review of odontogenic keratocysts and the behavior of recurrences. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006; 101:5-9.
2. Odell EW. *Cawson's Essentials of Oral Pathology and Oral Medicine*. 9th Ed. London: Elsevier; 2017. p.23-234
3. Mallya S, Lam EWN, editors. *White and Pharoah's Oral Radiology: Principles and Interpretation*. 8th Ed. London: Elsevier; 2019. p.391-3
4. Al-Moraissi EA, Kaur A, Gomez RS, Ellis E 3rd. Effectiveness of different treatments for odontogenic keratocyst: a network meta-analysis. *Int J Oral Maxillofac Surg* 2023; 52:32-43.
5. Mendes RA, Carvalho JFC, van der Waal I. Characterization and management of the keratocystic odontogenic tumor in relation to its histopathological and biological features. *Oral Oncol* 2010; 46:219-25.

6. Kimura T, Ohba S, Yoshimura H, Fujita S, Imamura Y, Kitagawa Y, *et al.* Keratocystic odontogenic tumor arising at the mandibular ramus with an impacted tooth: a case report and mimic lesions. *Cranio* 2016; 34:58-63.
7. Tsukamoto G, Makino T, Kikuchi T, *et al.* A comparative study of odontogenic keratocysts associated with and not associated with an impacted mandibular third molar. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002; 94:272-5.
8. Caixeta Guimarães A, Dutra de Cassia Ferreira Santos M, Machado de Carvalho G, Takahiro Chone C, Nizam Pfeilsticker L. Giant keratocystic odontogenic tumor: three cases and literature review. *Iran J Otorhinolaryngol* 2013; 25:245-52
9. Sembronio S, Albiero AM, Zerman N, Costa F, Politi M. Endoscopically assisted enucleation and curettage of large mandibular odontogenic keratocyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; 107:193-6.
10. Pahlevan R, Keyhanlou F, Fazel S, Shahsavari F. Keratocystic odontogenic tumor associated with impacted maxillary third molar extending to the antrum: A challenging diagnosis. *Hum. Pathol.: Case Rep* 2019;15:59-63.
11. Cakur B, Miloglu O, Yolcu U, Göregen M, Gürsan N. Keratocystic odontogenic tumor invading the right maxillary sinus: a case report. *J Oral Sci* 2008; 50:345-9.
12. Enislidis G, Fock N, Sulzbacher I, Ewers R. Conservative treatment of large cystic lesions of the mandible: a prospective study of the effect of decompression. *Br J Oral Maxillofac Surg* 2004; 42:546-50.
13. Dammer R, Niederdellmann H, Dammer P, Nuebler-Moritz M. Conservative or radical treatment of keratocysts: a retrospective review. *Br J Oral Maxillofac Surg* 1997; 35:46-8.
14. Chye CH, Singh B. Rapid cystic development in relation with an impacted lower third molar: a case report. *Ann Acad Med Singapore* 2005; 34:130-3.
15. Poonja P, Maryam H, Ratnakar H, Rithul P, Majumder B, Varghese A, *et al.* Odontogenic keratocyst with horizontal impaction of third molar. *Arch Med Health Sci* 2021; 9:113.
16. Pitak-Arnop P, Chainé A, Oprean N, Dhanuthai K, Bertrand JC, Bertolus C. Management of odontogenic keratocysts of the jaws: a ten-year experience with 120 consecutive lesions. *J Craniomaxillofac Surg* 2010; 38:358-64.
17. Gupta G, Gupta D K, Gupta P, Rana K S, Chandra N. An Unusual Odontogenic Keratocyst Associated with Third Molar Crypt Near Condyle in an Adolescent: A Case Report. *J Res Dent Maxillofac Sci* 2023; 8:57-61.
18. Tan B, Yan TS, Shermin L, Teck KC, Yoke PC, Goh C, Balakrishnan A. Malignant transformation of keratocystic odontogenic tumor: Two case reports. *American Journal of Otolaryngology-Head and Neck Medicine and Surgery* 2013; 34(4): 357-61.
19. Arshad F. Syndromic odontogenic keratocyst: A case report and review of literature. *J Int Soc Prev Community Dent* 2016; 6(1):84-8.
20. Kumar NN, Padmashree S, Jyotsna TR, Shastry SP. Gorlin-Goltz syndrome: A rare case report. *Contemp Clin Dent* 2018; 9(3):478-83.