Open Access Library Journal
2026, Volume 13, e14955
ISSN Online: 2333-9721
ISSN Print: 2333-9705

Mandibular Odontogenic Keratocyst in a Young Patient: A Case Report and Literature Review

Nadia El Haiba, Nadia El Wadidi, Ziad Farih, Mokrane Khazana, Youssef Naji

Department of Surgical Odontology, Mohammed VI Center for Dental Care, Mohammed VI Faculty of Dental Medicine, Mohammed VI University of Health Sciences (UM6SS), Mohammed VI Foundation for Health Sciences, Casablanca, Morocco
Email: nelhaiba@um6ss.ma

How to cite this paper: El Haiba, N., El Wadidi, N., Farih, Z., Khazana, M. and Naji, Y. (2026) Mandibular Odontogenic Keratocyst in a Young Patient: A Case Report and Literature Review. *Open Access Library Journal*, **13**: e14955.
https://doi.org/10.4236/oalib.1114955

Received: January 29, 2026
Accepted: March 8, 2026
Published: March 11, 2026

Abstract

Odontogenic keratocysts are frequent cysts of the jaws, representing 10% - 20% of odontogenic cysts. They develop from the dental lamina or its remnants and mainly affect young adults, with a slight male predominance. The mandible, particularly the angle and molar region, is the most frequent site. We report the case of an 18-year-old female presenting with left mandibular pain. Three-dimensional imaging allowed precise evaluation of the lesion's extent and its anatomical relationships. Management consisted of surgical enucleation with extraction of the associated tooth. Histopathological examination confirmed the diagnosis of an odontogenic keratocyst. Prolonged follow-up, essential to detect possible recurrence, was instituted. This case illustrates the importance of early diagnosis, 3D imaging, and rigorous follow-up in the management of odontogenic keratocysts.

Subject Areas

Oral and Maxillofacial Surgery

Keywords

Odontogenic Keratocyst, CBCT Imaging, Surgical Enucleation, Mandibular Cyst, Recurrence Prevention, Gorlin-Goltz Syndrome

1. Introduction

The odontogenic keratocyst, previously referred to as the keratocystic odontogenic tumor, was reclassified in 2017 by the World Health Organization (WHO) as an odontogenic keratocyst due to its cystic rather than neoplastic behavior [1]. It accounts for approximately 10% - 20% of all odontogenic cysts and occurs mainly in young individuals, with a peak incidence between the second and third

decades of life [2] [3].

The mandible is the most common site, particularly the angle and molar regions. Association with an impacted tooth is frequent, reported in 25% - 40% of cases [3]. The odontogenic keratocyst is often asymptomatic and discovered incidentally during routine radiographic examinations. However, symptoms such as pain, swelling, or infection may occur in the case of a large or complicated lesion [2]. Radiographic imaging, particularly three-dimensional CBCT, allows a precise assessment of the lesion's extent, cortical integrity, and relationship with the inferior alveolar nerve, which is essential for treatment planning [4].

2. Case Report

2.1. Medical History

An 18-year-old female patient presented with left mandibular pain (**Figure 1**). She reported no medical or surgical history and had no known allergies. She consumes alcohol occasionally. The patient described spontaneous pain in the left mandible, aggravated by mastication, which had been present for one week. Partial relief was obtained after a course of corticosteroid therapy (Cotipred for 3 days) combined with antibiotic therapy (Augmentin for 4 days).

Figure 1. Exo-buccal view: facial symmetry preserved, no visible swelling or abnormalities.

2.2. Clinical Examination

Extraoral examination revealed facial symmetry, no palpable lymphadenopathy, and a slight click in the left temporomandibular joint noticeable on mouth opening. Intraoral examination revealed the clinical absence of tooth 38. The mucosa adjacent to tooth 37 showed mild inflammation and was tender on palpation (**Figure 2**). Tooth 26 had a temporary zinc oxide-eugenol (ZOE) dressing. Tooth 36 was restored with a composite filling. Teeth 27 and 37 showed occlusal fissure infiltration. The vitality test of tooth 37 was positive, whereas the percussion test was negative.

2.3. Complementary Investigations

The panoramic radiograph (**Figure 3**) shows a well-defined radiolucent lesion

with clear, regular margins, homogeneous and unilocular in appearance, extending from the distal aspect of tooth 37 to the mesial aspect of tooth 38. The lesion has a rounded morphology, measuring approximately 25 mm in width and 40 mm in length. Tooth 38 is displaced distally and presents two immature, divergent roots, located in close proximity to the inferior alveolar nerve.

Reference panoramic reconstruction (CBCT) (Figure 4) showed progressive bucco-lingual extension.

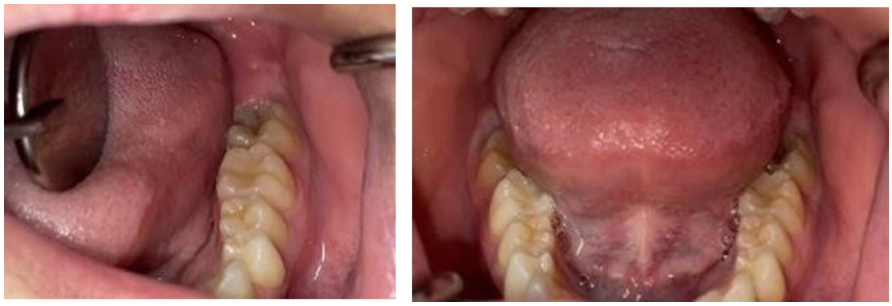

Figure 2. Intraoral view of the left mandibular half, affected region; the mucosa is slightly inflamed on the distal aspect of tooth 37.

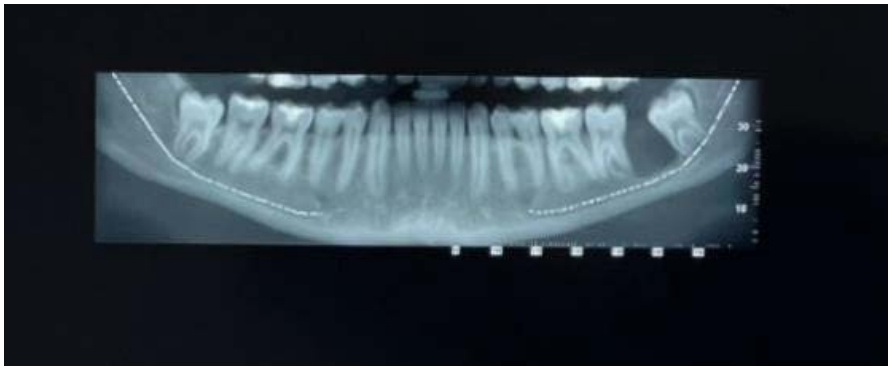

Figure 3. Panoramic view of the mandible showing the radiolucent lesion.

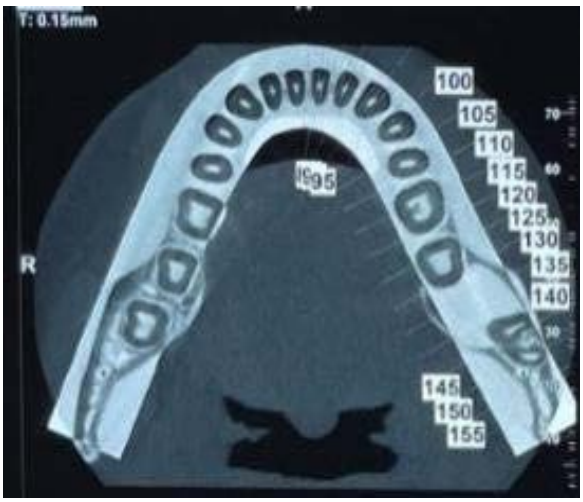

Figure 4. Reference panoramic reconstruction (CBCT) showing progressive bucco-lingual extension.

Oblique coronal reconstructions (Figure 5) revealed that the lesion arises at the level of the roots of tooth 37, with approximate dimensions of 20 mm in the vertical direction and 15–18 mm in the buccolingual direction, and is associated with a breach of the lingual cortical plate. It is in close relation to the inferior alveolar nerve.

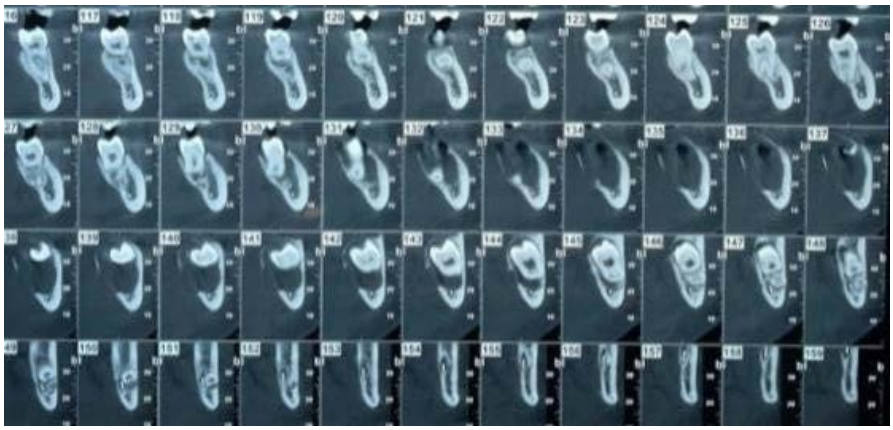

Figure 5. Oblique coronal reconstructions showing the lesion in relation to tooth 37 and the inferior alveolar nerve.

2.4. Histopathological Diagnosis

Histopathological examination (Figure 6) confirmed the diagnosis of an odontogenic keratocyst, revealing thin, regular, parakeratinized stratified squamous epithelium, associated with minimally inflamed connective tissue.

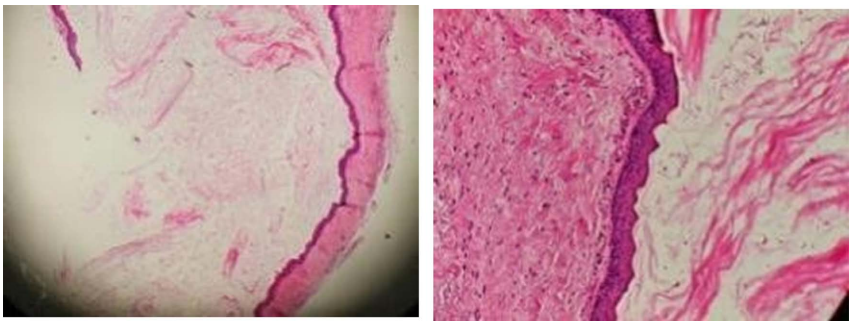

Figure 6. Histopathological section: (a) ×4 magnification, Hematoxylin-eosin staining; (b) ×40 magnification, Hematoxylin-eosin staining.

2.5. Treatment

The treatment consisted of complete enucleation of the cyst combined with extraction of tooth 38 under local and regional anesthesia (Figures 7-10).

Two-week and seven-week postoperative follow-ups showed satisfactory mucosal healing and initial bone reossification (Figure 11). At 12 months, panoramic imaging confirmed complete bone consolidation (Figure 12). Tooth 37 remained vital and asymptomatic throughout the entire follow-up period, including at the 12-month assessment, confirming the absence of pulpal compromise despite the

Figure 7. Full-thickness mucoperiosteal flap.

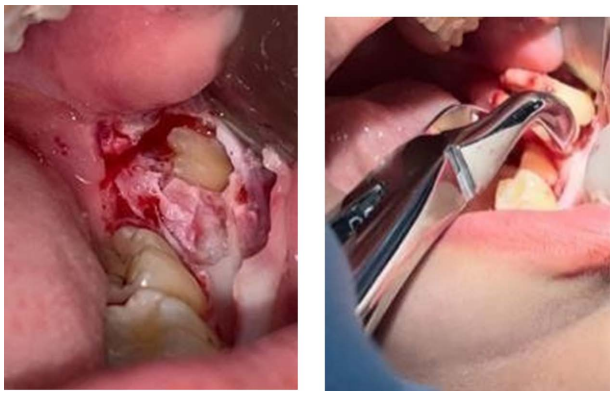

Figure 8. Du Surgical extraction of tooth 38.

Figure 9. Complete cyst enucleation performed without rupture of the cystic cavity.

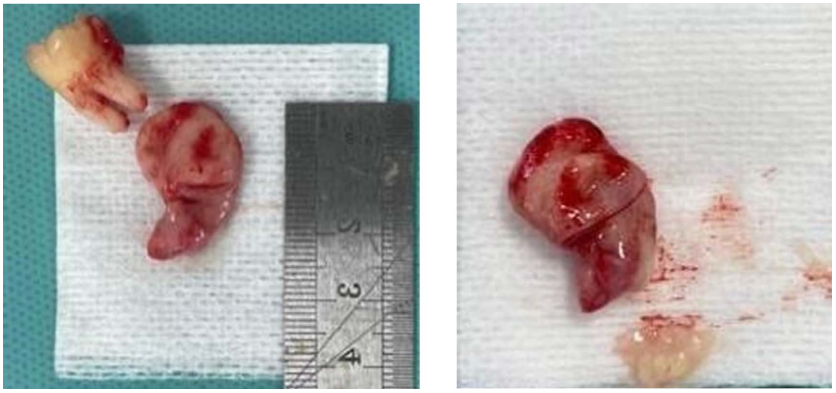

Figure 10. Surgical specimen showing tooth 38 and the lesion, measuring 2.5 cm.

Figure 11. Postoperative follow-up at 1.5 months and 12 months to assess mucosal healing and bone reossification.

Figure 12. Postoperative follow-up panoramic radiograph showing satisfactory bone healing and no evidence of recurrence.

proximity of the enucleation to the apical area of this tooth. The patient had no sensory deficits, confirming preservation of the inferior alveolar nerve.

3. Discussion

The odontogenic keratocyst (OKC), as defined in the 2017 WHO classification, accounts for approximately 10% - 20% of all odontogenic cysts and is distinguished by a unique biological behavior characterized by rapid growth and a high recurrence rate, setting it apart from more common odontogenic cysts [1] [5]. It preferentially occurs in young individuals, with a peak incidence between the second and third decades of life, corresponding to the age of the patient in this case, highlighting the importance of early management [2] [3].

Topographically, more than 60% - 80% of OKCs are located in the mandible, particularly in the angle and molar regions, where bone density, the presence of impacted teeth, and mechanical stress favor their development and explain the mandibular predominance of these lesions [6]. Association with an impacted

tooth is observed in 25% - 40% of cases, as illustrated by our patient presenting with an impacted tooth 38, reinforcing the link between tooth impaction and OKC development [7].

Clinically, OKCs are often asymptomatic and discovered incidentally during routine radiographic examinations. However, they may present with localized pain, progressive swelling, tooth mobility, or secondary infection, particularly when the lesion becomes large or causes cortical perforation, conditions that can complicate management [6] [8]. In our case, mandibular pain was the primary reason for consultation, illustrating the variability of OKC clinical presentations [8].

Radiographically, OKCs typically appear as well-defined radiolucent lesions, either unilocular or multilocular, with smooth or scalloped borders. They tend to displace the inferior alveolar canal without causing significant root resorption, a characteristic radiologic feature [6]. In our patient, CBCT imaging allowed precise evaluation of the lesion's three-dimensional extent, lingual cortical perforation, and close relationship with the inferior alveolar nerve, highlighting the importance of 3D imaging in surgical planning and neurovascular structure preservation, especially to anticipate potential neurological complications [4]. No root resorption was observed, confirming the typical OKC pattern of displacement rather than destruction of adjacent roots [6].

A fundamental aspect to address at this stage concerns the potential association of OKCs with **Gorlin-Goltz syndrome** (nevoid basal cell carcinoma syndrome, NBCCS), a rare autosomal dominant condition caused by a germline mutation of the *PTCH1* tumor suppressor gene, implicated in the Hedgehog signaling pathway [9] [10]. This syndrome is classically characterized by a triad consisting of multiple basal cell carcinomas appearing as early as adolescence, multiple and recurrent OKCs of the jaws present in 75% to 90% of affected individuals, and skeletal anomalies with ectopic calcifications, most notably calcification of the falx cerebri [9] [10]. OKCs occurring in the context of Gorlin-Goltz syndrome are generally multiple, bilateral, and early-onset, with a markedly higher recurrence rate compared to sporadic forms, underscoring the clinical importance of syndromic screening in any young patient presenting with an OKC [9] [10].

From a diagnostic standpoint, the criteria established by Kimonis *et al.* (1997) distinguish between major and minor criteria [10]. Major criteria include: two or more basal cell carcinomas (or one occurring before the age of 20), histologically confirmed odontogenic keratocysts of the jaws, three or more palmoplantar pits, calcification of the falx cerebri, rib or vertebral anomalies, and a first-degree family history of the syndrome. Minor criteria encompass macrocephaly, congenital malformations such as cleft lip or palate, polydactyly or ocular anomalies, specific radiological findings including bridging of the sella turcica, flame-shaped vertebral anomalies and hand or foot bone abnormalities, ovarian fibromas, and desmoplastic medulloblastomas. The diagnosis is confirmed in the presence of two major criteria, or one major criterion combined with two minor criteria [10].

Radiologically, OKCs associated with Gorlin-Goltz syndrome are often multiple, bilateral, and characterised by early recurrence. Panoramic radiography and CBCT play an essential role in their detection, surveillance, and surgical planning. Calcification of the falx cerebri, visible on skull radiographs or computed tomography, constitutes one of the major radiological criteria of the syndrome and should be systematically sought when the clinical context raises suspicion [9] [10]. The presence of multiple jaw cysts in a young patient must therefore always prompt a thorough search for associated syndromic signs, both clinically and radiographically. At the molecular level, PTCH1 gene mutations lead to constitutive activation of the Hedgehog signaling pathway, promoting uncontrolled cell proliferation and explaining the aggressive biological behaviour and high recurrence potential of OKCs in this syndromic context. This molecular understanding has guided the development of targeted therapeutic strategies, including vismodegib, a Hedgehog pathway inhibitor evaluated in severe or recurrent cases of Gorlin-Goltz syndrome [9] [11]. In the present case, the patient was systematically assessed for clinical signs of Gorlin-Goltz syndrome, including multiple basal cell carcinomas, skeletal anomalies, calcification of the falx cerebri, and a family history of the syndrome. No such features were identified, and syndromic association was therefore ruled out.

Nevertheless, given the patient's young age and the inherent recurrence potential of OKCs, continued vigilance during long-term follow-up remains warranted [9] [10].

The differential diagnosis primarily includes dentigerous cysts, paradental cysts, and unicystic ameloblastomas, which must be considered to avoid inappropriate treatment [2] [7]. The definitive diagnosis relies on histopathological examination, revealing thin, regular, parakeratinized stratified squamous epithelium, associated with minimally inflamed connective tissue and sometimes the presence of satellite cysts, typical morphological features of OKCs and recognized risk factors for recurrence [12].

Definitive treatment consists of complete enucleation of the lesion, generally combined with extraction of the associated impacted tooth to reduce recurrence risk, as recommended by most authors [6] [13] [14]. Despite this, the recurrence rate remains high, ranging from 15% - 30% or more in the absence of adjunctive measures, justifying long-term, rigorous follow-up [14] [15]. Adjunctive techniques, such as peripheral ostectomy, application of Carnoy's solution, or cryotherapy, are recommended in the presence of satellite cysts or marked connective tissue inflammation to limit recurrence [6] [14].

In our case, complete enucleation with extraction of tooth 38 was performed according to current recommendations. Adjunctive therapy was not employed because histopathological examination revealed no satellite cysts and only minimal connective tissue inflammation, which are the primary risk factors for recurrence justifying their use [6] [14]. In the absence of these histological risk factors, the standard protocol of enucleation combined with tooth extraction was deemed sufficient, consistent with established guidelines [2] [3] [13]. For large lesions, a con-

servative decompression approach may be considered to reduce the risk of bone fracture and facilitate secondary enucleation, a strategy validated by several clinical studies [8] [16].

Postoperative follow-up includes a two-week evaluation to assess mucosal healing and absence of early complications, a six-to-eight-week assessment to evaluate initial bone reossification and mandibular functional recovery, a 12-month follow-up to confirm bone consolidation and optimal healing through clinical and radiographic examination, and long-term follow-up of five years or more to detect early recurrences and ensure complete functional and aesthetic restoration, reflecting the necessity of prolonged, rigorous monitoring [14] [15].

In our patient, this follow-up protocol was applied and proved fully consistent with the clinical evolution. Postoperative assessments at two weeks and seven weeks confirmed satisfactory mucosal healing and progressive bone reossification, while the 12-month panoramic radiograph demonstrated complete bone consolidation with no evidence of recurrence. Throughout the entire follow-up period, tooth 37 remained vital and asymptomatic, confirming the absence of pulpal compromise despite the proximity of the surgical site to its apex. The patient presented no sensory deficit, attesting to preservation of the inferior alveolar nerve. Importantly, given the unilocular nature of the lesion, its limited buccolingual extent, the absence of satellite cysts on histopathological examination, and the complete preservation of tooth 37, no additional restorative or complementary treatment was deemed necessary. The topography of the lesion and the favourable clinical situation thus allowed a conservative approach without further intervention beyond standard follow-up [6] [14] [15].

At the molecular level, recent studies highlight the involvement of the Hedgehog signaling pathway and PTCH1 gene mutations, particularly in Gorlin-Goltz syndrome, explaining the aggressive behavior and recurrence potential of OKCs and guiding the development of targeted therapeutic strategies, paving the way for personalized treatments [9]. In the present case, the patient was systematically assessed for clinical signs of Gorlin-Goltz syndrome (nevoid basal cell carcinoma syndrome), including multiple basal cell carcinomas, skeletal anomalies, calcification of the falx cerebri, and a family history of the syndrome. No such features were identified, and syndromic association was therefore ruled out.

Nevertheless, given the patient's young age, continued vigilance during long-term follow-up remains warranted [9].

This case illustrates a typical presentation of a mandibular OKC in a young patient, emphasizing the importance of early diagnosis, meticulous surgical planning based on three-dimensional imaging, systematic screening for Gorlin-Goltz syndrome, and standardized, long-term follow-up to minimize recurrence risk and ensure optimal functional and aesthetic outcomes, in line with literature recommendations [9] [10] [14] [15].

4. Conclusion

The odontogenic keratocyst is characterized by aggressiveness and a higher recur-

rence rate than other jaw cysts. For this reason, it has long been considered a benign cystic neoplasm rather than a simple odontogenic cyst. Diagnosis relies on the combination of clinical, radiological, and histopathological findings [6]. Although surgery remains the mainstay of treatment, therapeutic strategies are varied and remain debated in the literature. Systematic evaluation for Gorlin-Goltz syndrome is recommended in all young patients presenting with odontogenic keratocysts, given the potential syndromic context and its implications for long-term multidisciplinary management. Prolonged follow-up is essential to detect and manage any recurrence effectively.

Acknowledgements

The authors would like to thank the Department of Oral Surgery and the Dental Care Center of UM6SS for their support in this case management.

Conflicts of Interest

The authors declare no conflicts of interest.

References

[1] Regezi, J.A., Sciubba, J.J. and Jordan, R.C. (2017) Oral Pathology: Clinical Pathologic Correlations. 7th Edition, Elsevier.

[2] Pogrel, M.A. (2003) Decompression and Marsupialization as a Treatment for the Odontogenic Keratocyst. *Oral and Maxillofacial Surgery Clinics of North America*, **15**, 415-427. https://doi.org/10.1016/s1042-3699(03)00038-4

[3] Shear, M. and Speight, P. (2007) Cysts of the Oral and Maxillofacial Regions. Blackwell. https://doi.org/10.1002/9780470759769

[4] MacDonald-Jankowski, D.S. (2011) Keratocystic Odontogenic Tumour: Systematic Review. *Dentomaxillofacial Radiology*, **40**, 1-23. https://doi.org/10.1259/dmfr/29949053

[5] Philipsen, H.P. (2005) Keratocystic Odontogenic Tumour. In: Barnes, L., Eveson, J.W., Reichart, P. and Sidransky, D., Ed., *WHO Classification of Tumours. Pathology and Genetics of Head and Neck Tumours*, IARC Press, 306-307.

[6] Stoelinga, P.J.W. (2001) Long-Term Follow-Up on Keratocysts Treated According to a Defined Protocol. *International Journal of Oral and Maxillofacial Surgery*, **30**, 14-25. https://doi.org/10.1054/ijom.2000.0027

[7] Kaczmarzyk, T., Mojsa, I. and Stypulkowska, J. (2012) A Systematic Review of the Recurrence Rate for Keratocystic Odontogenic Tumour in Relation to Treatment Modalities. *International Journal of Oral and Maxillofacial Surgery*, **41**, 756-767. https://doi.org/10.1016/j.ijom.2012.02.008

[8] Anthony Pogrel, M. and Jordan, R.C.K. (2004) Marsupialization as a Definitive Treatment for the Odontogenic Keratocyst. *Journal of Oral and Maxillofacial Surgery*, **62**, 651-655. https://doi.org/10.1016/j.joms.2003.08.029

[9] Gorlin, R.J. (1995) Nevoid Basal Cell Carcinoma Syndrome. *Dermatologic Clinics*, **13**, 113-125. https://doi.org/10.1016/s0733-8635(18)30114-1

[10] Kimonis, V.E., Goldstein, A.M., Pastakia, B., Yang, M.L., Kase, R., DiGiovanna, J.J., *et al.* (1997) Clinical Manifestations in 105 Persons with Nevoid Basal Cell Carcinoma

Syndrome. *American Journal of Medical Genetics*, **69**, 299-308. https://doi.org/10.1002/(sici)1096-8628(19970331)69:3<299::aid-ajmg16>3.0.co;2-m

[11] Tang, J.Y., Mackay-Wiggan, J.M., Aszterbaum, M., Yauch, R.L., Lindgren, J., Chang, K., *et al.* (2012) Inhibiting the Hedgehog Pathway in Patients with the Basal-Cell Nevus Syndrome. *New England Journal of Medicine*, **366**, 2180-2188. https://doi.org/10.1056/nejmoa1113538

[12] Li, T.J. (2011) The Odontogenic Keratocyst. *Journal of Dental Research*, **90**, 133-142. https://doi.org/10.1177/0022034510379016

[13] Blanas, N., Freund, B., Schwartz, M. and Furst, I.M. (2000) Systematic Review of the Treatment and Prognosis of the Odontogenic Keratocyst. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*, **90**, 553-558. https://doi.org/10.1067/moe.2000.110814

[14] Pogrel, M.A. (2013) The Keratocystic Odontogenic Tumor. *Oral and Maxillofacial Surgery Clinics of North America*, **25**, 21-30. https://doi.org/10.1016/j.coms.2012.11.003

[15] Daley, T.D., Wysocki, G.P. and Pringle, G.A. (1994) Recurrence of Odontogenic Keratocysts: A Study of 137 Cases. *Oral Surgery, Oral Medicine, Oral Pathology*, **77**, 112-118.

[16] Toller, P. (1967) The Treatment of Large Keratocysts by Marsupialization. *British Journal of Oral Surgery*, **5**, 145-155.