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Presentation and management of syndromic and non-syndromic patients with multiple odontogenic keratocysts

Omer Sefvan Janjua [a], Ramish Tariq [b], Muhammad Usman Khalid [a], Sana Mehmood Qureshi [c], Kamran Ali [d,*]

[a] Oral and Maxillofacial Surgeon, Allied Hospital/Faisalabad Medical University, Faisalabad, Pakistan
[b] BDS, Postgraduate Resident Oral and Maxillofacial Surgery, Allied Hospital/FMU, Faisalabad, Pakistan
[c] Oral Pathology, Faisalabad Medical University, Faisalabad, Pakistan
[d] Oral Surgery, Qatar University, College of Dental Medicine, Visiting Professor Plymouth University, Devon, Qatar

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ABSTRACT

Objective: This case series reports the clinical presentation and management of multiple odontogenic keratocysts (OKCs) in patients with Gorlin Goltz syndrome and in non-syndromic patients.
Material and methods: Eight patients presented with features of multiple odontogenic keratocyst at department of Oral and Maxillofacial Surgery. The diagnosis was confirmed following clinical, radiographic and histopathological examination. Initially the diagnosis of OKC was confirmed on an incisional biopsy. Major and minor criteria were followed for the diagnosis of Gorlin Goltz syndrome. Smaller cysts in all patients were enucleated and for larger cysts marsupialization was planned either alone or followed by enucleation. Patients were followed at 1 month, 3 months, 6 months and yearly interval to check for bone healing and recurrences.
Results: Association of Gorlin Goltz syndrome was identified in four patients all of whom were males with age range of 12–37 years. Among four patients that had non-syndromic OKCs, three were female and one was male with age range of 09–50 years; two patients had familial non-syndromic multiple OKCs. In syndromic patients, multiple OKCs, recognized manifestations of Gorlin Goltz syndrome were identified with variable frequency: calcification of falx cerebri and chest deformity (100%); macrocephaly with frontal bossing (100%); hypertelorism (75%); basal cell carcinoma (25%); pectus deformity with flame shaped hands and feet (25%) and syndactyly (50%). Palmar or plantar pits, cleft lip or palate, ovarian fibroma and medulloblastoma was not identified in any patient. In patients with non-syndromic multiple OKCs mandible was more commonly involved than maxilla.
Conclusion: Patients with multiple OKCs, should be evaluated thoroughly and basal cell carcinomatous lesions should be ruled out. Meticulous follow up is vital as Gorlin Goltz syndrome is associated with malignancies and OKCs may be the first manifestation of this syndrome. Given the fact that OKCs associated with this syndrome have higher rate of recurrence than the isolated OKCs, long term follow up is warranted.

* Corresponding author.
E-mail addresses: osj1982@hotmail.com (O.S. Janjua), ramisht@ymail.com (R. Tariq), drusmankhalid82@hotmail.com (M.U. Khalid), sana.mehmood@outlook.com (S.M. Qureshi), ali.kamran@qu.edu.qa (K. Ali).

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1. Introduction

Odontogenic keratocyst (OKC) is the third most common cystic lesion of the oral cavity [1]. It was previously called as primordial cyst or Keratocystic odontogenic tumor (KCOT) but in 2017 it was renamed as odontogenic cyst [2]. OKC is a developmental cyst and originates from the rest cells of dental lamina [3]. It is a benign, locally destructive lesion and may be unicystic or multicystic [4].

The incidence of OKC is 10–20% of all the odontogenic cyst [2]. Most of the cases are reported during first to eighth decade of life with peak reported in 30 years of age and more commonly occurring in males [5]. Lining of the cyst is parakaratinized stratified squamous epithelium with a thickness of 5–10 layers of cells. The basal layer consists of cuboidal or columnar cells. Epithelium connective tissue interface is flat and has high mitotic activity resulting in daughter cells [6].

Mandible is more commonly involved as compared to the maxilla. The most common site in mandible is the molar region and the ramus whereas in maxilla anterior region is the favorite site of OKC followed by third molar region. It may be associated with the lateral root or the periapical region of the teeth or with the crown of the tooth [5]. OKCs are associated with medullary expansion and buccolingual expansion may not be visible until the lesions become advanced. The recurrence rate of OKCs is high may range from 25% to 60%. This may be attributed to a thin, friable epithelial lining which may rupture during removal, presence of daughter cysts. Moreover, new OKC lesions may develop from remnants of dental lamina [7].

Treatment options consists of conservative and surgical management i.e., enucleation, marsupialization, with or without peripheral procedures like curettage, peripheral ostectomy, Carnoy's solution, electrocautery, cryotherapy and resection [8].

Gorlin Coltz syndrome (GCS) is also known as nevoid basal cell carcinoma syndrome (NBCCS) and is the most recognized syndrome associated with multiple OKCs [9]. Less commonly oral-facial-digital syndrome, Noonan syndrome, Ehlers-Danlos syndrome and Simpson-Golabi-Behmel syndrome may also present with multiple OKCs [10]. Patient with multiple OKCs are reported to have a higher recurrence rate [1]. Multiple OKC not associated with a ny syndrome are reported rarely [3].

This case series reports management of patients presenting with multiple OKCs with and without associated Gorlin Goltz syndrome.

2. Methods

. Eight patients presented to the Oral and Maxillofacial Surgery department, Allied Hospital Faisalabad from November 2017 to March 2021 and included three females and five males. The common presenting symptoms included swelling, pus discharge, or pain in at least one quadrant of the oral cavity; one patient also presented with pathological fracture of the mandible. All patients were initially investigated with orthopantomogram (OPG). Multiple well defined, radiolucent lesions involving the maxilla and mandible were observed in all patients (Fig. 1). The lesions in all patients presented as a combination of multilocular and unilocular radiolucencies.

Clinical examination and investigations according to the protocol described by Lo Muzio [10] were followed to rule out the syndromic association of multiple cystic lesions. Frontal bossing, macrocephaly (>55 cm), hypertelorism with increased intercanthal distance (>36 cm), palmar and plantar pits along with aspiration of the cystic lesion was done on clinical examination. Out of eight patients, four were suspected to be associated with Gorlin Goltz syndrome (GGS). These patients were further advised CBCT, brain CT, and radiographs for chest, spine, hand and feet. These patients were evaluated by dermatology, neurology, ophthalmology and otorhinolaryngology specialists.

Initially incisional biopsies of the cystic lesion were performed. Presence of a parakaratinized stratified squamous epithelium with 5–6 cell layer thickness, basal palisading, a thin connective tissue capsule and luminal content of cheesy proteinaceous material were used to confirm the diagnosis of OKC (Fig. 2).

Four patients with features of GCS, were investigated further regarding any family history (FH) of cysts and tumors or other features of syndrome and first-degree relatives examined for any positive findings. Syndromic association was not identified in four patients

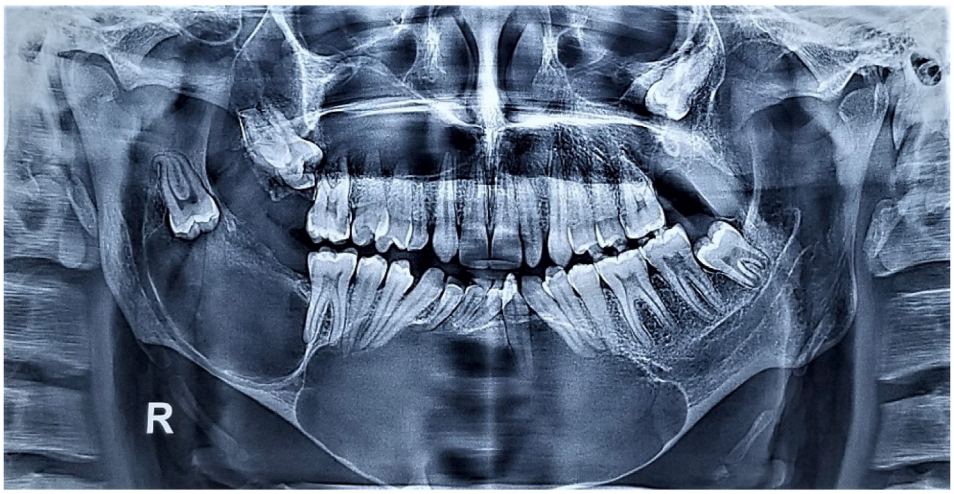

Fig. 1. Orthopantomogram (OPG) showing multiple cystic lesions in the maxilla and the mandible.

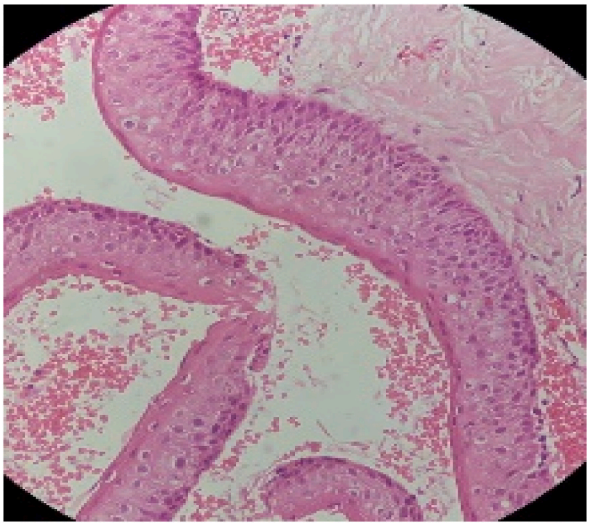

Fig. 2. Histopathology showing parakaratinized squamous epithelium at low power (10) magnification with H&E staining.

and they did not require additional investigations.

Enucleation with peripheral ostectomy was planned for cysts less than 3 cm in size. For larger cysts marsupialization alone or followed by enucleation was planned. Associated impacted teeth were extracted and root canal therapies were done for teeth that were deemed to be restorable. Smaller cystic cavities were closed primarily and antiseptic bismuth iodoform paraffin paste (BIPP) dressing was placed in larger cystic cavities that were changed every week. Two patients with non-syndromic familial multiple OKCs refused treatment. The remaining the patients were followed closely. OPG was repeated at 03-month, 06-month, 09 month and at 12 months. Bone healing was monitored radiographically for 12 months with serial radiographs at 03 months intervals.

Ethics approval for the study was gained from the institutional review board (IRB) for publication of this case series study.

3. Results

Among four patients who were diagnosed as having GGS, all were male with an age range of 12–35 years. All the patients were diagnosed following the major and the minor criteria of GGS (see Table 1). Considering the major criteria, 100% had multiple OKC involving the maxilla and mandible. The most common site of cyst was mandibular body followed by ramus and then the posterior maxilla. Maxillary cyst was symptomatic only in one patient. 100%of patients had calcification of falx cerebri (Fig. 3). Chest deformity consisting of bifid ribs, were present in 100% of patients with scoliosis in 25% of patient (Fig. 4). First degree relative was involved in 25% of patient. Basal cell carcinoma (BCC) of chest was found in only one patient and he was referred to general surgery for the treatment of BCC (Fig. 5). Palmar or plantar pits were not identified in any patient in this group.

Considering the minor criteria for NBCCS, macrocephaly was present in 100% of cases (Fig. 6). Frontal bossing (Fig. 7) and coarse face was present in 4 (100%) of them with hypertelorism present in 3 (75%) cases. Pectus deformity was present in 1 (25%) patient, flame shaped hands and feet in 1 (25%) patient and syndactyly was noted in 2 (50%) patients (Fig. 8). Cleft lip or palate, ovarian fibroma or medulloblastoma were not identified in any patient.

Among four patients that had non-syndromic OKCs, three were female and one was male with an age range of 9–50 years. Among them two patients had familial non-syndromic multiple OKCs. One patient had three cysts in the mandible and one in the maxilla, one patient had four in the mandible and one cyst in maxilla, one had two cysts in the maxilla and one in the mandible and the other patient was found to have one cyst in the maxilla and two in the mandible. One patient was asymptomatic and multiple cystic lesions were identified incidentallyon radiographic investigations. Rest of the three patients were asymptomatic for the maxillary cysts and presented to us with symptomatic mandibular cysts. Two patients who had familial non-syndromic OKC had cysts associated with impacted teeth. One patient had impacted right maxillary third molar and left maxillary second molar associated with cyst. The other patient had cyst associated with impacted left maxillary second molar, left mandibular canine, right mandibular second premolar, right mandibular canine and lateral incisor and had retained deciduous right mandibular lateral incisor, canine and second molar.

Enucleation with peripheral ostectomy was done for small cystic lesions and marsupialization alone or followed by enucleation was done for larger cysts in all patients. Out of four syndromic patient one patient had recurrence after 14 months of follow up and had again undergone enucleation of the cysts. One patient was lost to follow up. Two patients are still under treatment and had not shown any recurrence so far.

Out of four non-syndromic patients, two patients with familial non-syndromic OKC refused to treatment. Rest of the two patients had not shown any recurrence till date.

Table 1
Presentation and management of odontogenic keratocysts.

Sr. No.	Age	Gender	Syndromic/ Non-syndromic	Major criteria [1–25]	Minor criteria	No. of cysts (=N)	Location of cysts	Management	Follow-up
1	12yrs	Male	Yes	Multiple OKCs Calcification of falx cerebri Bifid ribs	Macrocephaly Frontal bossing Coarse face Hypertelorism Syndactyly	2	Anterior mandible (n = 1) Anterior maxilla (n = 1)	Marginal resection Enucleation	No recurrence since 1 year
2	25yrs	Male	Yes	Multiple OKCs Calcification of falx cerebri Bifid ribs	Macrocephaly Frontal bossing Coarse face Hypertelorism Syndactyly	4	Anterior mandible (n = 1) Posterior mandible (n = 1) Posterior maxilla (n = 2)	Marsupialization Marsupialization Marsupialization and enucleation	No recurrence for 1 year
3	32yrs	Male	Yes	Multiple OKCs Calcification of falx cerebri Bifid ribs First degree relative was involved BCC of chest	Macrocephaly Frontal bossing Coarse face	5	Posterior mandible (n = 2) Posterior maxilla (n = 2) Anterior maxilla (n = 1)	Enucleation Enucleation Enucleation	Recurrence after 08 months of surgery
4	35yrs	Male	Yes	Multiple OKCs Calcification of falx cerebri Bifid ribs, scoliosis	Macrocephaly Frontal bossing Coarse face Hypertelorism Pectus deformity Flame shaped hands and feet	4	Posterior mandible (n = 2) Anterior mandible (n = 1) Anterior maxilla (n = 1)	Marsupialization Marsupialization Enucleation	Did not show for follow up
5	9yrs	Female	No	–	–	4	Posterior mandible (n = 2) Anterior mandible (n = 1) Anterior maxilla (n = 1)	Enucleation Enucleation Enucleation	No recurrence since I year
6	13yrs	Female	No	–	–	5	Posterior mandible (n = 2) Anterior mandible (n = 2) Posterior maxilla (n = 1)	Refused treatment	–
7	24yrs	Female	No	–	–	4	Anterior mandible (n = 2) Posterior maxilla (n = 2)	Refused treatment	–
8	50yrs	Male	No	–	–	3	Anterior mandible (n = 1) Posterior mandible (n = 1) Anterior maxilla (n = 1)	Enucleation Marsupialization followed by enucleation	No recurrence for 1 year

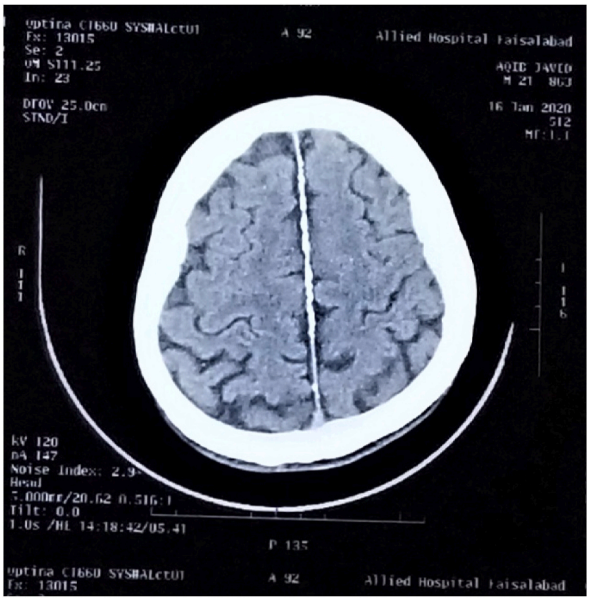

Fig. 3. Axial CT scan of brain showing calcification of falx cerebri.

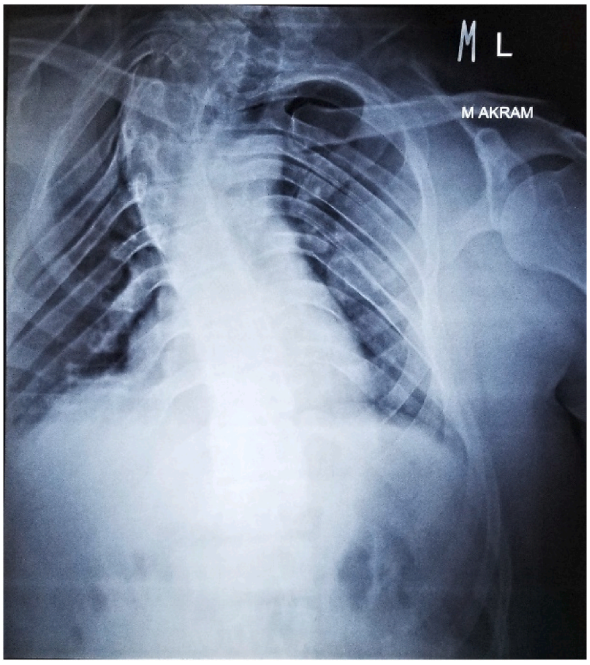

Fig. 4. Chest radiograph (PA view) showing scoliosis of spine.

4. Discussion

OKC was first described by Mikulicz in 1956. It is a developmental cyst arising from the overlying basal epithelium or from the rests of dental lamina [11]. OKC can occur in any age group with peak age reported from 10 to 40 years [3]. Reported ratio of occurrence of OKC in mandible and maxilla is 2:1 [11]. In line with the literature, mandible was involved more commonly in the present study in syndromic as well as non-syndromic cases. OKCs often present as the initial lesions of GCS followed by other symptoms at a later stage [2]. Patients with single OKC are mostly older in age as compared to multiple OKCs occurring in either syndromic or non-syndromic patients [12].

Small OKCs are often asymptomatic while larger cysts may present with swelling, pain or discharge [13]. Patients in the current case series presented to us with swelling, pain or discharge and one patient with pathological fracture of the mandible. Multiple OKCs

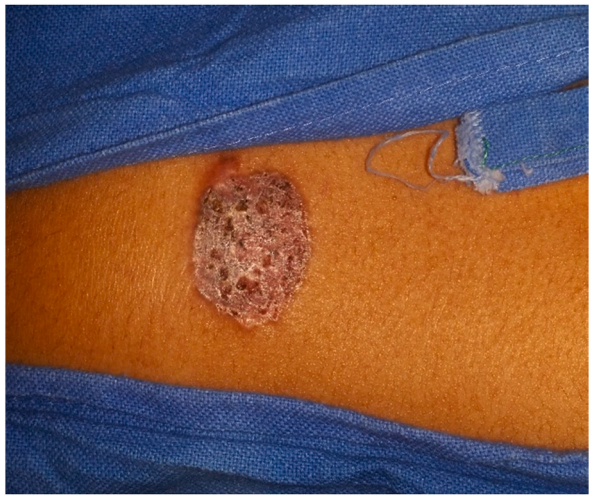

Fig. 5. Basal Cell Carcinoma on the anterior chest wall at the level of 7th rib.

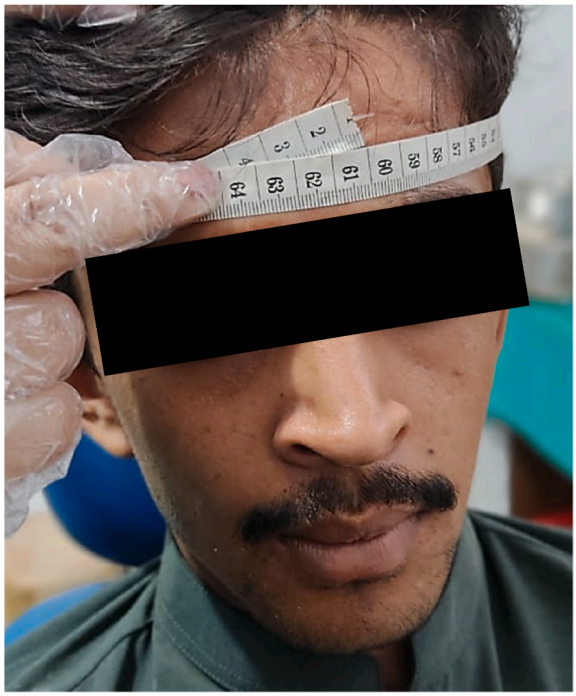

Fig. 6. Showing macrocephaly with enlarged head circumference.

are most commonly associated with GCS while non-syndromic cases often present with solitary lesions [3]. Kimonis et al. modified the criteria for the diagnosis of GCS. It has major and minor criteria. Diagnosis for GCS requires two major criteria, or one major and two minor criteria [14].

The major criteria are:

- Multiple BCCs or one occurring under the age of 20 years.
- OKCs of the jaws confirmed on histopathological examination.
- Palmar or plantar pits (three or more).
- Bilamellar calcification of the falx cerebri.
- Bifid, fused or markedly splayed ribs.
- First-degree relative with NBCCS.

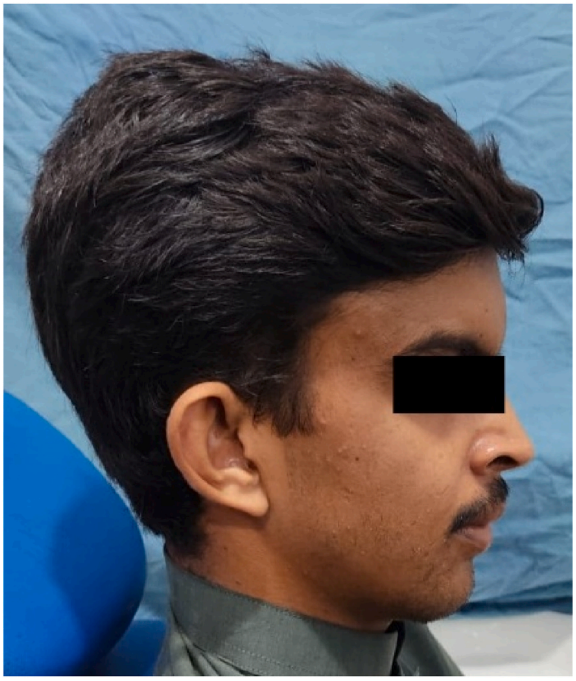

Fig. 7. Lateral view of the face showing frontal bossing.

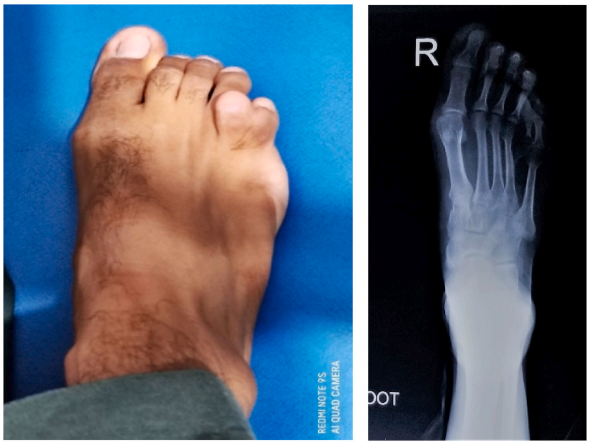

Fig. 8. Polydactyly of fifth metatarsal.

The minor criteria are:

- Macrocephaly
- Congenital malformation: cleft lip or palate, frontal bossing, coarse face, moderate or severe hypertelorism.
- Other skeletal abnormalities: Sprengel deformity, marked pectus deformity, marked syndactyly of the digits
- Radiological abnormalities: Bridging of the Sella turcica, vertebral anomalies such as hemivertebrae, fusion or elongation of the vertebral bodies, modeling defects of the hands and feet or flame shaped hands or feet.
- Ovarian fibroma.
- Medulloblastoma.

In our study four patients with GCS, one patient fulfilled 5 major criteria i.e., multiple OKCs, calcification of falx cerebri, bifid ribs, BCC, first degree relative was also affected and had 2 minor criteria i.e., frontal bossing and macrocephaly. The second patient had 3 major criteria i.e., multiple OKCs, calcification of falx cerebri, bifid ribs and had 4 minor criteria i.e., frontal bossing, macrocephaly, syndactyly and hypertelorism. The third patient had 3 major criteria i.e., multiple OKCs, calcification of falx cerebri, scoliosis and had 5 minor criteria i.e., frontal bossing, macrocephaly, hypertelorism, flame shaped hands and feet and marked pectus deformity. Finally,

the fourth patient had 3 major criteria i.e., multiple OKCs, calcification of falx cerebri, bifid ribs and had 4 minor criteria i.e., frontal bossing, macrocephaly, hypertelorism and syndactyly. Bomfin et al. and Balland et al. in their studies found the incidence of multiple OKCs in GCS from 62 to 100% [15,16]. Reported incidence of macrencephaly (>55 cm) is 5–80%, calcification of falx cerebri in 21.2–92%, hypertelorism is 6–78%, bifid rib is 16–58%, ovarian cysts in 25–50%, cleft lip and palate in 0–9%, in patients of NBCCS [17,18]. Although chromosomal mapping is advisable for patients with multiple OKCs, this facility was not available at the study center.

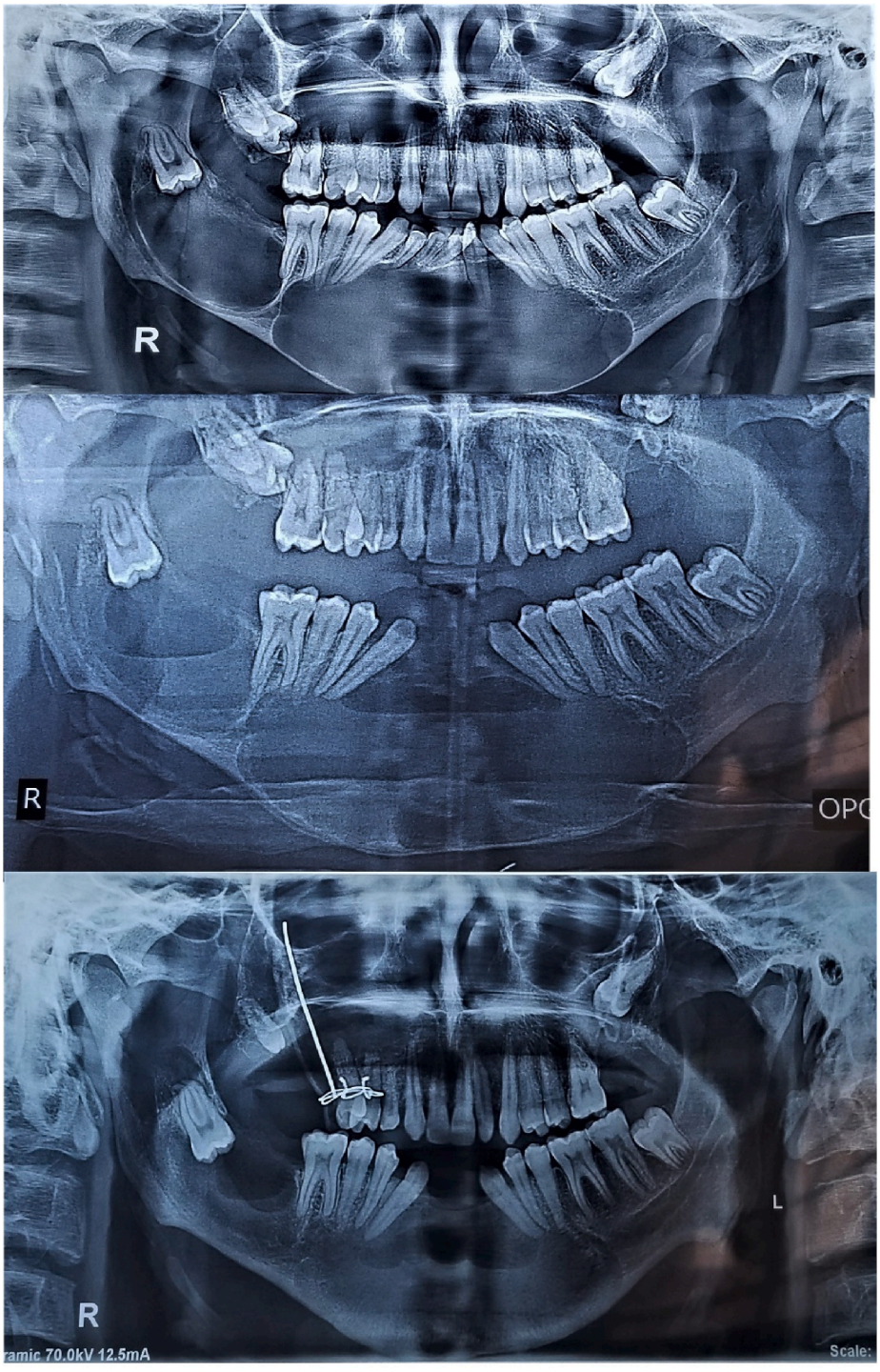

Fig. 9. (a) Preoperative OPG (b) postoperative OPG after 6 months of marsupialization (c) postoperative after 9 months of marsupialization.

Four patients in our study had multiple non-syndromic OKCs, constituting half of the cohort in this case series. Brannon in his study of 312 cases of OKC found that only 5.8% of cases had multiple OKCs without any associated syndrome [19]. Parikh and Sholapurkur et al. in their study reported cases of multiple OKCs that were not associated with any syndrome [20,21]. Habibi et al. and Bartake et al. also reported same results in their studies [6,22].

Out of four syndromic patients, one patient reported with recurrence of multiple OKCs, while one patient did not report back for follow-up. (Fig. 9). The patient with recurrence was re-treated with enucleation and peripheral ostectomy of the cysts. Out of four non-syndromic patients, two patients with familial multiple OKCs refused treatment, while the two treated cases showed no recurrence after 1 year follow up.

Previous literature has reported some differences between syndromic and non-syndromic OKCs. The mean age of presentation of OKCs in syndromic patients is 40 years and in non-syndromic is 10–30 years, with a predilection for females in syndromic and male in non-syndromic cases. Histopathological findings in non-syndromic OKCs report a thin epithelial lining, fewer mitotic figures and satellite cysts [12]. In the present study, syndromic patients had a mean of age of 29 years with range of 12–35 years and non-syndromic had an age range of 9–50 years. Male predominance was seen in syndromic patients and female in non-syndromic patients. Histopathology of OKCs did not identify any difference between the syndromic and non-syndromic cases.

The main limitation of this study is that genetic screening of patients was not undertaken due to resource constraints. GCS is transmitted as an autosomal dominant (AD) trait and the reported prevalence range from 56,000–164,000 [23]. Molecular genetics studies show that GCS involves mutations in the PTCH (Patched) gene found on chromosome arm 9q22.3, 9q31, and 1p32 [24,25]. Mutations in GCS underscore the need for undertaking genetic testing in the diagnostic work-up for patients with suspected syndromic involvement such as occurrence of multiple OKCs. This approach also provides an opportunity for identifying hereditary risks and providing genetic counseling to affected families.

5. Conclusion

Patients presenting with OKC should be investigated for the presence of additional OKCs at the time of presentation and also during the follow up. Presence of multiple OKCs warrants a thorough assessment to rule out syndromic association as OKCs may be the first and only manifestation of this syndrome. Given that OKCs associated with this syndrome have higher rate of recurrence than the isolated OKCs, long term follow-up of patients is also essential. Affected patients need support and counseling regarding risk of developing malignancies such as BCC and other complications.

Author contributions

OS Janjua conceptualised the study and carried out the data analysis. R Tariq, M U Khalid and S M Qureshi were responsible for data collection. K Ali was responsible for review and final drafting of the manuscript.

Declaration of competing interest

None of the authors have any conflicts of interest to declare.

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