CASE SERIES

Congenital granular cell epulis: Pattern of presentation in four Nigerian tertiary hospitals

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Abstract
This study aims to report seven cases of congenital granular cell epulis (CGCE) diagnosed in four teaching hospitals in Southwestern Nigeria over a period of 8 years. CGCE is a rare lesion that presents exclusively at birth. It is benign, but often presents an unsightly appearance, and may be associated with difficulty in feeding, thus early presentation of cases for treatment. The records of the oral and maxillofacial pathology departments of four Nigerian teaching hospitals were assessed for lesions with histological diagnosis of CGCE. Data inclusive of age, gender, anatomic site, and mode of treatment was retrieved and analyzed with SPSS version 21. Seven cases were identified from the four teaching hospitals. All cases occurred in females with an average period of presentation at 7 days after birth. Most of the cases (71.4%) occurred on the maxillary alveolar ridge. Six cases presented as a single pedunculated or sessile lesion, while in one case, three nodules were seen. Although spontaneous regression has been documented in literature, all our cases had surgical excision under local anesthesia without complications or recurrence. All our cases were diagnosed in females and 71.4% were on the maxillary alveolar ridge.

Keywords: Congenital epulis, granular, Nigeria

Introduction
Congenital granular cell epulis (CGCE) is a benign lesion mostly seen on the maxillary alveolar ridge of newborns. It was first reported by Neumann, in 1871, hence, the nomenclature Newman’s tumor.1 The third edition of the World Health Organization classification of head-and-neck tumors adopted the name “CGCE.”2 It is a rare lesion with estimated incidence of 0.0006%.3 Review of literature by Zuker and Buenchea, in 1993,4 revealed 167 reported cases within 122 years of its description, and recently, a review of cases seen globally by Singh et al. reported 156 cases.5 It occurs more on the maxilla than mandible in the ratio of 3:1 and affects more females than males in the ratio of 8:1.6 Occasionally, the lesion may occur on the mandibular alveolar ridge. The size of the lesion ranges from a few millimeters to as much as 6 cm.7 Its association with congenital anomalies such as transverse facial cleft, polydactyl, congenital goiter, and neurofibromatosis8 and its presence in an in vitro fertilized baby have also been documented.9

Although prenatal evidence of lesion on routine ultrasound of pregnant mother have been documented,10 diagnosis of CGCE is by histology, which presents as large sheets of polygonal or rounded cells with a centrally placed small dark basophilic nucleus with an abundant eosinophilic granular cytoplasm. Lesion is often overlain by parakeratinized stratified squamous epithelium with a high degree of vascularity. The treatment is surgical excision under general anesthesia or local anesthesia,11,12 excision under topical anesthesia in a small lesion of 1 cm has been reported.13 Follow-up post-excision has ranged from 3 months to 12 years without recurrence.14,15 The absence of malignant transformation and recurrence following surgical excision has been consistent with all documented cases. CGCE has been reported from Nigeria13-16 and other African countries.17,18 This study presents a case series of seven new cases from the archives of oral and maxillofacial pathology laboratories of some Nigerian institutions.

The aim of this study is to present the clinical and histologic features of seven new cases seen over 8 years, as well as present an update of knowledge of CGCE.
Materials and Methods

The records of oral and maxillofacial pathology departments in four teaching hospitals, in Southwestern Nigeria, from 2010 to 2018 were assessed for cases of lesions with histologic diagnosis of CGCE. Together, these are the foremost referral centers for histopathology analysis of oral and maxillofacial pathologies in Southwestern Nigeria. The glass slides along with clinical details including age at presentation, size of lesion, anatomic location, and treatment received were retrieved and reviewed by AA and SA; both agreed on the diagnoses.

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Seven new cases of CGCE were diagnosed in these centers from 2010 to 2018 [Table 1]; all occurred in females and were present at birth, average age of presentation was 7 days. Five (71.4%) of the seven cases occurred on the maxillary alveolar ridge, while 2 cases (28.6%) were observed on the mandibular alveolar ridge. Six of the eight cases were solitary while one was multiple. Six cases were pedunculated and lobulated, while one case was sessile [Figure 1].

Macroscopic view of CGCE often reveals an oval shaped firm tissue mass which may be single [Figure 2] or multiple. Microscopically, all the lesions were composed of large polygonal cells with abundant granular eosinophilic cytoplasm; nuclei were basophilic, round to oval, and central. Highly vascularized stroma and an overlying well-differentiated stratified squamous epithelium were seen in all cases [Figure 3a and b].

All the cases were treated by surgical excision under local anesthesia. Follow-up ranged from 1 week to 6 months without any evidence of recurrence.

Discussion

In this study, we have described seven new cases of congenital epulis seen in four Nigerian hospitals over an 8-year period, this corroborates the report by Singh et al., 2018,[5] in which a review of 156 cases revealed a majority (42.9%) diagnosed in Southeast Asia followed by India (20.5%). Others were diagnosed in Europe (17.3%), Malaysia (8.3%), the United States (7.7%), China (7.1%), and Brazil (6.4%). Sixteen (6.4%) of the 156 cases were diagnosed in sub-Saharan Africa, four of which were in Nigerians while the remaining three documented cases from Nigeria by Adenike et al., 2013,[17] were not captured in that series.

This study is a relatively large case series as only a few studies have described more than four cases in a case series in the scientific literature [Table 2].[20-23]

![Figure 1: Clinical picture of patient with lesion in situ](image1)

![Figure 2: Macroscopic view of excised lesion of congenital granular cell epulis](image2)

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex</th>
<th>Duration</th>
<th>Site</th>
<th>Size</th>
<th>Clinical description</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Female</td>
<td>10 days</td>
<td>Maxillary alveolar ridge</td>
<td>4 cm</td>
<td>Multiple pedunculated, hemorrhagic</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>2</td>
<td>Female</td>
<td>30 days</td>
<td>Maxillary alveolar ridge</td>
<td>1.5 cm</td>
<td>Pedunculated</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>3</td>
<td>Female</td>
<td>4 days</td>
<td>Maxillary alveolar ridge</td>
<td>1.3 cm</td>
<td>Smooth sessile</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>4</td>
<td>Female</td>
<td>3 days</td>
<td>Mandibular alveolar ridge</td>
<td>2 cm</td>
<td>Lobulated and pedunculated</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>5</td>
<td>Female</td>
<td>1 day</td>
<td>Maxillary alveolar ridge</td>
<td>4.5 cm</td>
<td>Encapsulated noodle</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>6</td>
<td>Female</td>
<td>3 days</td>
<td>Mandibular ridge</td>
<td>2.1 cm</td>
<td>Lobulated and pedunculated</td>
<td>Excision under local anesthesia</td>
</tr>
<tr>
<td>7</td>
<td>Female</td>
<td>3 days</td>
<td>Maxillary ridge</td>
<td>3 cm</td>
<td>Lobulated and pedunculated</td>
<td>Excision under local anesthesia</td>
</tr>
</tbody>
</table>
All of our cases occurred in females, a finding similar to some other studies. The reason for the female predominance has not been established, as hormonal influences of estrogen and progesterone have not been detected; however, endogenous maternal or fetal hormonal influences during pregnancy have been suggested as possible mechanism for the female predominance.

In this series, our patients presented at an average of 7 days similar to the report of Kupers et al., 2009. Although lesions were observed at birth by the parents, Jalil et al., 2015, reported an average age of 16 days at presentation (range 2–90 days). The reason for the delay in some of the cases was unknown; negative cultural attitudes are a possibility as observed in congenital anomalies such as cleft lip and natal teeth. This might have discouraged the parent from seeking immediate medical assistance. Maxillary presentation was more predominant in our series, which conforms to the predominant documented site. About 88.5% of the cases in the review by Singh et al., 2018, were maxillary lesions. The average size of the lesion in this present study was 2.75 cm, with the largest mass measuring 4.5 cm. This is similar to findings by Jalil et al., 2015, which reported a mean size of 2.2 cm, the largest of which was 4.0 cm. The mean size of the lesion in a systematic review of CGCG was of 2.9 ± 1.6 cm and lesions ranged from 0.3 cm to 8 cm. All cases in this series were surgically excised without complications. Studies on the origin of this lesion have remained inconclusive, despite immunohistochemical studies toward the identification of its histogenesis.

**Conclusion**

The clinical presentation of CGCE in Nigerian patients is typical of findings reported in literature and surgical excision is the treatment option employed.

**Clinical significance**

CGCE is a rare lesion present at birth most commonly seen in females and maxillary alveolar ridge.

**Acknowledgment**

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**References**