

Case Report





# Remission of congenital epulide - case report

#### **Abstract**

The lesion of congenital granular cells, also known as Neumann Tumor or Congenital Epulide Tumor, is a benign and rare congenital lesion, with incidence in newborns with predilection by the female gender and occurs predominantly in the anterior alveolar ridge of the maxilla. This lesion exhibits benign behavior and there are no reports of relapse in the literature. This report aims to describe the observed case of a newborn seven days old, melanoderma, presenting lesion of congenital granular cells in the gingival mucosa of the maxilla with difficulties in breastfeeding. The treatment consisted of observation and control, since there was a natural rupture of the lesion, without other complications and without recurrence, culminating with the natural return of the newborn to breastfeeding.

**Keywords:** congenital abnormalities, neoplasms, pediatric dentistry, newborn, prenatal diagnosis

Volume 9 Issue 6 - 2018

Milena Tavares de Carvalho, <sup>1</sup> Isadora Santana de Souza, <sup>2</sup> Patrícia Maria Coelho, <sup>3</sup> Clarissa Teles Fagundes, <sup>4</sup> Edite Novais Borges Pinchemel, <sup>5</sup> Loy Carvalho de Castro <sup>5</sup>

Department of Pediatric Dentistry, São Leopoldo Mandic

School, Professor at FAINOR, Brazil

<sup>2</sup>Department of Dentistry, FAINOR, Brazil

<sup>3</sup>Master in Radiology from PUC-MG, Professor at FAINOR, Brazil

<sup>4</sup>Professor at FAINOR, Brazil

<sup>5</sup>Bahia State University of Southwestern Bahia (UESB), Brazil

Correspondence: Milena Tavares de Carvalho, Department of Pediatric Dentistry, São Leopoldo Mandic School, Professor at FAINOR, Rua Ulisses do Prado, 18, CEP 45028-662, Vitória da Conquista, Brazil, Tel (55) 77 981309339, Email carvaho.mile@gmail.com

Received: July 26, 2017 | Published: November 19, 2018

## Introduction

Dentistry for newborns is willing to provide a lower rate of dental problems to children's development, presenting more condition of proper oral health and parents more aware of the relevance of prevention. Attention is now focused on early care, beginning before the first year of life, when children of a few days or months are taken to the dental office to have the first educational recommendations and diagnosis of anomaly or alteration in the oral cavity, solving limitations such as difficulty in breast-feeding. Therefore, the need to be able to distinguish and diagnose correctly and safely, anomalies in early stages, initiating treatment.1 Tumors of the oral cavity are not common among newborns. Congenital epulide of the newborn, evaluated as a tumor of congenital granular cells, is a rare gingival neoplasia that compromises the alveolar ridge of the newborn. Its clinical exposure is usually a single tumor located in the anterior alveolar ridge of the maxilla. The lesion may be sessile or pedunculate, with normal or gingival red coloration, of firm consistency, smooth and lobed surface. The caliber of the lesion may change, its existence may restrict the complete closure of the baby's mouth and most of the tumor presents benign without further development after birth, indicating absence of metastasis and recommending treatment as early as possible.<sup>2</sup> Congenital granular cell lesion, although rare, is simple to diagnose due to the typical manifestation in the alveolar ridge of the newborn, presenting as a generally single-variant submucosal mass, predominantly female. Its occurrence is sporadic, with no associated genetic load there are reports of relapse in the literature. The recommended treatment is surgical excision, especially when there is compromise during breastfeeding, swallowing and breathing of the newborn.<sup>3</sup> It is ideal that it be diagnosed and treated early because of the patient's premature age. Delayed diagnosis can compromise adjacent structures, inhibiting their rehabilitation. Through a multidisciplinary project and follow-up during the trans and postoperative period, the patient has good postoperative conditions and immediate esthetics, besides allowing oral speech and food intake. The objective of this study was to report a case of Congenital Epulide with remission of symptomatology and natural resolution of the cause without surgical intervention.

### Case report

This report describes the case of a newborn seven days old, female, melanoderma, born in Vitória da Conquista, Bahia - Brazil. In the clinical examination, a lesion was observed in the mucosa of the upper border with a nodular aspect and pedunculated base, presenting small lesion, of pink color in the gingival mucosa of the maxilla (Figure 1). The lesion presented a smooth surface with an erythematous base, measuring approximately 1cm in diameter (Figure 2). The infant showed difficulties in breastfeeding, interrupted sleep and frequent crying associated with a painful process, according to the mother's report. The initial diagnosis was Congenital Epulide after clinical examination, and the responsible child was informed that an incisional biopsy would be performed for the purposes of diagnostic confirmation and treatment (Figure 3). However, there was no need for surgical intervention, since on the same day of the visit there was a natural rupture of the lesion and remission of symptoms. The return of breastfeeding was gradually achieved with total wound healing, with no relapses or other complications, demonstrating that remission of symptomatology and natural resolution of the cause is possible, the newborn was then followed up with observation and control. The term of Free and Informed Consent was signed by the mother before the consultation.





Figure I Clinical aspect of the lesion.



Figure 2 View of the nodular lesion in the border region.



Figure 3 Remission of the lesion after three months.

#### **Discussion**

The objective of the present report is based on exposing a case of natural remission of Congenital Epulide, with favorable prognosis. <sup>2,3,7</sup> Lucas RP et al 2014 reported that the Congenital Epulide was first described by Neumann in 1871 as a lesion affecting newborns, typically located in the alveolar ridge, specifically in the area of the incisors and canines. Thus, infants with this lesion may develop difficulties during the breathing process and/or feeding. <sup>1,4,5,6,7</sup> José BR

et al., 2015 describe the Congenital Epulide as a rare pathology, with an incidence eight times higher for females than for males and three times higher for maxilla than for mandible.4-7 About 200 cases have been described in the literature up to now.6 Epulide is indicated as a benign tumor, consisting of eosinophilic cells, containing granules in the cytoplasm and mainly present in the alveolar region.8 Dionline BP et al.<sup>10</sup> describe that the clinical manifestation is based on a lobular or ovoid, sessile or pedunculated edema covered by a surface of smooth mucosa.10 The diagnosis is made clinically after birth and is confirmed histologically. Although the histogenesis of the Congenital Epulide is not certain, it is believed to be a non-neoplastic lesion. 9,10 According to Van der Waal, the immunohistochemical findings can be explained by the fact that they include several different cell populations such as fibroblasts, histiocytes, perineural cells and perivascular cells among others. The Congenital Epulide presents microscopic features composed of fusiform and ovoid cells without granular cytoplasm. 11,12 Galileu BG et al. 2005 report that hormonal aspects are questionable during gestation.<sup>13</sup> Basílio AM et al., 2010 has shown many theories to explain histogenesis and one of the most accepted theories is the possible influence of intrauterine ovarian hormones of the fetus. 12,14 For prenatal diagnosis, it is suggested to perform estrogen receptor and progesterone influence tests,9 but this idea is still an experience and currently most diagnoses of Congenital Epulide are determined after birth.3 Muhaizan WM et al. 2016 mention that the differential diagnosis is made through histological examination, which characterizes the proliferation in regular layers of granulosa cells. 15 Danny KCW et al., 2016 report that granule cells in the Epulide are specifically S-100 negative protein.<sup>15</sup> Among the differential diagnosis we can mention the Bohn nodules, Epstein pearls, mucocele, eruption cyst, neuroectodermal tumor and Abrikossoff tumor.5 Histogenesis is contested in different studies and suggests several origins for Epulide such as histiocytes, smooth muscle and primitive mesenchymal cells. 3,4,6,16-19 The Epulide belongs to the mesenchymal benign tumor, presenting reports of spontaneous regression and absence of local recurrence after excision.<sup>3,6</sup> The most important lesions to differentiate from Congenital Epulide are oral teratoma-epignathus<sup>14</sup> and neuroectodermal melanocytic tumor of childhood. 9,19,20 The diagnosis is basically clinical by the revelation of an intraoral mass. However, the disclosure can be made by an obstetric ultrasound device from the 36th week of gestation. 10,21-23 Computed tomography or magnetic resonance imaging is useful for demonstrating the size and planning of differential treatment, when the lesion obstructs and impedes the feeding or breathing process of the infants.<sup>23,24</sup> The result after surgical removal is often favorable and there were no reports of recurrence or irregularities in the future dentition of the newborns.<sup>3,10,25-27</sup> The treatment of Epulide has two routes, surgical and non-surgical with spontaneous regression of the lesion, as in the case in question. 19,23,28 Surgical excision is indicated and there are no reports of recurrence. The spontaneous regression of the lesion is rare, 12,16,19,28,29 given the importance of reporting the case described. Many authors seek to understand the pathology, although the etiology of the Congenital Epulide remains uncertain and remains unknown. The present work reports a rare case of Congenital Epulid where it presents remission of the symptomatology, without the necessity of the surgical intervention, without relapse of the same.

### Conclusion

The present report showed a case of Congenital Epulide in a newborn with natural rupture without the need of surgical intervention, contributing to the complete remission of the symptoms.

# **Aknowledgements**

None.

#### Conflict of intrest

The authors declares that there is no conflcit of interest.

#### References

- Beatriz HES, Sinara HG, Mariana ND, et al. Characteristics of the Oral Cavity of Newborn Babies. *Pesq Bras Odontoped Clin Integr*. 2012;12(1):89–92.
- João GSP, Humberto TG, José LP, et al. A rare case of congenital epulis of the newborn with multiple lesions. Rev odonto ciênc. 2008;23(4):403–406.
- Marcelo RS, Karina ML, Gabriel HF, et al. Congenital granular cell lesion: a case report. Rev Cir Traumatol Buco-Maxilo-fac. 2010;10(1):35–38.
- Camilla RP, Renato M. Dental strategy with immediate palatal obturator in a patient with melanotic neuroectodermal tumor of infancy: case report. Rev bras odontol. 2015;72:(1-2):4-9.
- Ana SB, Soledad BC, Ramón RG, et al. Anesthetic Management of Congenital Epulis. Rev Col Anest. 2011;39:405–408.
- Adriane SS, Márcia RDC, Ana CDM, et al. Congenital epulis with autoresolution: case report. RGO, Rev Gaúch Odontol. 2014;62(3):315–18.
- Patricia RB, Vera CA, José BR, et al. Multiple Congenital Granular Cell Epulis: Case Report and Immunohistochemical Profile with Emphasis on Vascularization. Case Reports in Dentistry. 2015.
- Hideharu K, Jouji N, Yoshihiko M, et al. A Case of Congenital Granular Cell Epulis in the Maxillary Anterior Ridge: A Study of Cell Proliferation Using Immunohistological Staining. *J Maxillofac Oral* Surg. 2013;12(3):333–337.
- 9. Deepak S, Srinivas M, Tejo P, et al. Twin congenital epulis in the alveolar ridge of the maxilla and mandible in a newborn: a rare and interesting case. *BMJ Case Rep.* 2014.
- Dionline BP, Bruna RB, Jerúsia OI, et al. Congenital Granular Cell Lesion: A Case Report. Case Reports in Clinical Medicine. 2014;3:566–569.
- Andreas CP, Michail DN, Vasileios FK, et al. Spindle Cell Epulis in an 8-Month-Old Child: A Histologic Variant of Congenital Granular Cell Epulis. *Head and Neck Pathol*. 2012;6(4):467–470.
- Mark SL, Thomas JF, Alba G. Congenital Granular Cell Epulis Immunohistochemical and Ultrastructural Observation. Acta Dermatoven. 2007;3.

- Renato AA, Galileu BG, Cecília LP, et al. Congenital epulis with autoresolution: case report. RGO, Rev gaúch odontol. 2005;53(3):206–209.
- Marcelo RS, Karina ML, Gabriel HF, et al. Congenital granular cell lesion: case report. 2010;10(1):35–38.
- Wong DK, Ramli R, Muhaizan WM, et al. Congenital epulis: A rare benign tumour. Med J Malaysia. 2016;71(5):300–301.
- Aparna HG, Jayanth BS, Shashidara R, et al. Congenital Epulis in a newborn: A case report, Immunoprofiling and review of literature. *Ethiop J Health Sci*. 2014;24(4):359–362.
- 17. Maria TLM, Juárez LG. Diagnóstico de tumores de cara y cuello en el feto. Revista Peruana de Ginecología y Obstetricia. 2013;(4)16.
- Wong DK, Ramli R, Muhaizan WM, et al. Congenital epulis: A rare benign tumour. Med J Malaysia. 2016;71(5):300–301.
- David B, Graham R. Congenital Epulis: A Case Report and Estimation of Incidence. *International Journal of Otolaryngology*. 2009.
- Federico BJ. Epúlide congénito del recién nacido, una sorpresa para Neonatólogo Congenital epulis of the newborn, a surprise to the neonatologist. Rev Nac. 2015;7(2):37–39.
- Fister P, Volavsek M, Novosel SM, et al. A newborn baby with a tumor protruding from the mouth. *Acta Dermatoven*. 2007;16(3):128–130.
- Kristie S, Jacqueline S, Brendan M, Robert Magotti, Ron Benzie, Daniel. October 2006. 72;8.
- Federico BJ. Epúlide congénito del recién nacido, una sorpresa para

   Neonatólogo Congenital epulis of the newborn, a surprise to the neonatologist. Rev Nac (Itauguá). 2015;7(2):37–39.
- Fister P, Volavsek M, Novosel SM, et al. A newborn baby with a tumor protruding from the mouth. Acta Dermatoven APA. 2007;16(3):128–130.
- 25. Maria TLM, Juárez LG. Diagnóstico de tumores de cara y cuello en el feto. *Rev Latin Perinat*. 2013;16(4).
- Komla G, Komlan AMA, Harefetéguéna B, et al. Congenital giant epulis obstructing oral cavity: newborn emergency. African Medical Journal. 2014;17:195.
- Fister P, Volavsek M, Novosel SM, et al. A newborn baby with a tumor protruding from the mouth. Acta Dermatoven APA. 2007;16(3):128–130.
- Monika D, Anna K, Adriana GK, et al. Multiple congenital epulide (congenital granular cell tumours) in the newborn: a case report and review of literature. PJP 1 2011: Layout. 2011.
- McGuire TP, Gomes PP, Freilich MM, et al. Congenital Epulis: A Surprise in the Neonate. 2006;72(8):747–750.