Granular cell tumor in female neonate: a case report

Tumor de células granulares em neonato do sexo feminino: relato de caso

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ABSTRACT

The granular cell tumor (GCT) is a neoplasm of connective tissue which rarely occurs in children. This case report aims to describe and discuss the presentation of this tumor in a female neonate. Compared to previous literature, the case report demonstrated a rare form of presentation of this tumor in an unusual age group.

Key words: granular cell tumor; neonate; mouth neoplasm.

INTRODUCTION

Granular cell tumor (GCT) is an uncommon neoplasm of connective tissue that can occur anywhere in the body, but most cases affect the tongue. Apparently, there is a predilection for females, and blacks are more affected than whites\(^1,2\). The lesion occurs most frequently between the fourth and sixth decades of life, and is rare in children. The usual clinical presentation is an asymptomatic solitary nodule on the anterior portion of the tongue. Most GCTs present as a benign, not ulcerated and usually painless nodule, with insidious onset and slow growth rate. In such cases, complete surgical excision is usually curative.

CASE REPORT

A female neonate, 38 weeks and 3 days, 3845 g, 51.5 cm height, pink skin and Apgar 9/10. She was hydrated, suckling at mother’s breast vividly, with normal defecation and normal reflexes present. Normal colored mucous membranes; no heart
defects or respiratory disorders; normal vital signs; there were no other comorbidities. Upon examination of the oral cavity, a pedicle formation inserted in the right upper gingival line of about 1 cm was noted, with no loss of continuity and with no palate deformity. She was transferred to the neonatal intensive care unit (ICU) for preoperative preparation. The procedure was carried out with no complications; the patient presented normal vital signs. On the second day of life, she performed the neonatal hearing and tongue screening test, which showed normal results. She was discharged from the neonatal ICU on the same day.

The excised lesion was referred for anatomopathological analysis measuring 1 × 0.8 × 0.6 cm and presenting a brownish-white nodular appearance; GCT was evidenced. Subsequently, the diagnosis was complemented with an immunohistochemical study: positive for CD68 and negative for cytokeratin and S100. The findings favored the diagnosis of congenital granular cell epulis (Figures 1 to 3).

**DISCUSSION**

GCT is a benign tumor, with a predilection for the oral cavity, mainly for the tongue. It is uncommon in the gingival region, as in the patient case study. Recurrence rates of benign lesions are 2%-8%, even when the resection margins do not show tumor evidence(1). However, about 1% to 2% of histologically benign tumors can metastasize, via hematogenous route; the most common sites are bones, regional lymph nodes, peritoneal cavity and lungs(3). It is typically found as a solitary tumor, which needs to be differentiated from a squamous cell carcinoma lesion. In many cases, it appears as a yellowish-white, non-encapsulated nodule, less than 2 cm in size, with no ulceration or pain.

Malignant tumors are rare. They are usually larger than 5 cm and locally destructive, causing symptoms of obstruction, hemorrhage, ulceration and secondary infection. In addition, they show rapid growth, with local recurrence and distant metastases. The case of newborn in the study presented characteristics of benign nodules: small, non-ulcerated and painless. For patients with nodules with this aspect, the complete surgical excision is usually curative. The diagnosis is performed by histopathological analysis (HP) and immunohistochemistry (IHC). HP usually shows pseudoepitheliomatous hyperplasia, and it is necessary to rule out features that indicate malignancy(1, 4, 5). Nevertheless, the distinction between a benign and a malignant tumor is difficult; therefore IHC is indicated for definitive diagnosis.

Most studies show positivity for S100 and CD68; in the case studied, there was positivity only for CD68. Some studies suggest that markers may be negative when pseudoepitheliomatous hyperplasia of the epithelium is present(6). Patient’s IHC findings,
compared with other studies, favored the diagnosis of congenital granular cell epulis (GCT of the newborn). The recommended treatment is surgical resection, and radiotherapy can be used in the recurrent cases.

REFERENCES

1. Brannon RB, Anand PM. Oral granular cell tumors: an analysis of 10 new pediatric and adolescent cases and a review of the literature. J Clin Pediatr Dent. 2004 Fall; 29(1): 69-74. PubMed PMID: 15554407.

2. Richmond AM, Larosa FG, Said S. Granular cell tumor presenting in the scrotum of a pediatric patient: a case report and review of the literature [Internet]. J Med Case Rep. 2016; 10: 161. Available at: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4893259/. [Accessed on: 4 Jun, 2016].

3. van de Loo S; Thunnissen E, Postmus P , van der Waal I. Granular cell tumor of the oral cavity; a case series including a case of metachronous occurrence in the tongue and the lung [Internet]. Med Oral Patol Oral Cir Bucal. 2015; 20(1): e30-33. Available at: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4320418/. [Accessed on: 1 Jan, 2015].

4. Nasser H, Danforth RD Jr., Sunbuli M, Dimitrijevic O. Malignant granular cell tumor: case report with a novel karyotype and review of the literature. Ann Diagn Pathol. 2010; 14(4): 273-8. PubMed PMID: 20637434.

5. Fanburg-Smith JC, Meis-Kindblom JM, Fante R, Kindblom LG. Malignant granular cell tumor of soft tissue. Am J Surg Pathol. 1998; 22(7): 779-94. PubMed PMID: 9669341.

6. Le BH, Boyer PJ, Lewis JE, Kapadia SB. Granular cell tumor: immunohistochemical assessment of inhibin-a, protein gene product 9.5, S100 protein, and Ki-67 proliferative index with clinical correlation. Arch Pathol Lab Med. 2004; 128: 771-5. PubMed PMID: 15214825.