

Oral Subcutaneous Midline Leiomyomatous Hamartoma Presenting as Congenital Incisive Papilla Overgrowth in a Toddler

Abstract

Congenital soft-tissue tumors of oral cavity are mostly hyperplastic and benign in nature. This article presents an unusual case of congenital subcutaneous hamartoma of incisive papilla in a 2-year-old female child causing feeding and breathing difficulty. Total excisional biopsy was done under local anesthesia. Histopathology of tissue in reticulin-stained slide showed the presence of immature muscle fibers whereas Masson's trichrome stain revealed collagen fibers and smooth muscles confirming the diagnosis of oral midline subcutaneous smooth muscle (leiomyomatous) hamartoma of incisive papilla. It is important for dental professionals to be aware of this oral lesion present from birth mimicking overgrowth of incisive papilla, by its presentation, differential diagnosis, histopathology, and management.

Keywords: Hamartoma, hypertrophy, incisive papilla

Introduction

Congenital epulis, the rare benign soft-tissue tumors, derived from Greek word meaning “on the gum present with an aggressive appearance with large size, causing swelling, and difficulties during the breastfeeding.”^[1,2] Any exophytic, asymptomatic soft-tissue tumor in infants, unifocal/multifocal of the mucogingival zone of the anterior labial gingiva, should be considered in the differential diagnosis of the following nosologic entities: granular cell tumor, gingival cyst of the newborn, hamartoma, choristoma, vascular malformations, melanotic neuroectodermal tumor of infancy, oral teratoma, and rhabdomyosarcoma. This paper reports a rare case of congenital oral midline subcutaneous smooth muscle (leiomyomatous) hamartoma of incisive papilla in a rare presentation as solid tubular overgrowth of incisive papilla.^[3]

Case Report

A 2-year-old female child accompanied by her mother referred to the Department of Pediatric Dentistry with chief complaint of a solid tubular overgrowth of incisive papillary region which leads to difficulty in feeding and respiration. Medical history was normal, and the mother

reported normal, uncomplicated full-term pregnancy. Intraoral examination revealed growth on incisive papilla, i.e., palatal aspects of primary maxillary central incisors. This growth was present as a small nodule at birth which progressively increased in size extending to 2–2.5 cm below the incisal edge of maxillary central incisors [Figure 1]. The soft-tissue examination of the lesion revealed a solitary (2–2.5 cm × 0.5 cm × 0.5 cm) firm, painless, pedunculated growth on the incisive papilla. Soft-tissue examination revealed normal oral mucosa, gingiva, tongue, and floor of the mouth. Total excisional biopsy was done under local anesthesia, which was sent for histopathological examination in 10% formalin solution. Uneventful postoperative healing was observed, and no recurrence was reported.

Histopathological examination

Hematoxylin- and eosin-stained paraffin-based sections (5 µm thick) showed a parakeratinized stratified squamous epithelium with long and thick rete ridges. Underlining connective tissue stroma showed irregular and interlacing bundles of collagen fibers and numerous muscle fiber bundles. Plump- and spindle-shaped fibroblasts along with proliferating nerve bundles and endothelial cell-lined blood vessels underneath the epithelial surface

How to cite this article: Loomba A, Garg S, Dhindsa A, Kaur H, Jain N, Dhindsa P. Oral subcutaneous midline leiomyomatous hamartoma presenting as congenital incisive papilla overgrowth in a toddler. *Contemp Clin Dent* 2017;8:148-50.

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Access this article online

Website:

www.contempclindent.org

DOI: 10.4103/ccd.ccd_249_17

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Figure 1: (a and b) Growth attached to incisive papilla between deciduous maxillary central incisors. (c) Tissue received after excisional biopsy

with extravasated red blood corpuscles [Figure 2]. Reticulin-stained slide which showed the presence of immature muscle fibers whereas Masson's trichrome stain revealed collagen fibers and smooth muscles. Histopathological findings confirmed the diagnosis of oral midline subcutaneous smooth muscle (leiomyomatous) hamartoma of incisive papilla.

Discussion

Hamartoma is an abnormal proliferation of mature tissues, composed of elements that are normally found in the normal location for the tissue in which it develops, often with one predominating element. Hamartomatous muscle fibers remain in a dormant phase of cell cycle. This accords with differences between neoplasia and dysgenesis. Similar lesion like choristoma is a histologically normal tissue proliferation of a type that is normally not found in the anatomic site. The diagnosis of a lesion would, therefore, depend on its anatomic site. The present case appeared clinically as a simple soft-tissue mass present at birth; therefore, diagnosis of congenital epulis was made. Excisional biopsy was done under local anesthesia as feeding and respiration problems were encountered. Due to rare site of the involvement of incisive papilla, clinical diagnosis was confirmed by histopathological examination. Reticulin-stained slide showed the presence of immature muscle fibers whereas Masson's trichrome stain revealed immature collagen fibers and smooth muscles confirmed the diagnosis of oral leiomyomatous hamartoma mimicking hypertrophy of incisive papilla. The component elements of hamartoma are not foreign to that organ or tissue. Intraoral smooth muscle hamartomas of midline in palate are very rare and tend to occur at sites where fusion of processes and prominences occurs in developing embryo [Table 1].

Differential diagnosis for congenital epulis includes hemangioma, fibroma, rhabdomyoma, rhabdomyosarcoma, lymphangioma, sarcomas, hamartomas, teratoma, and

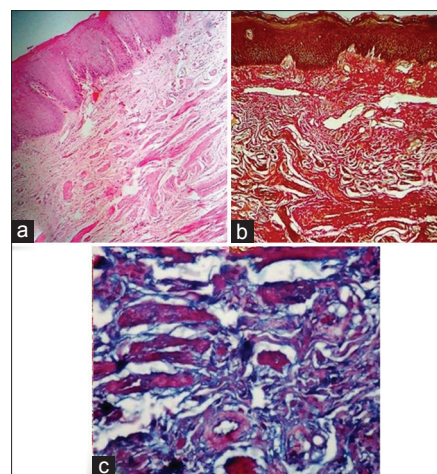


Figure 2: (a) H and E stained section showing parakeratinized stratified squamous epithelium with irregular and interlacing bundles of collagen fibers along with numerous muscle fiber bundles in connective tissue stroma. (b) Reticulin-stained section showing immature collagen fibers. (c) Masson's trichrome-stained section showing collagen fibers (blue) and smooth muscles (red)

granular cell tumor.^[4] This is defined as a cohesive tumor-like mass consisting of normal cells in abnormal location. Choristomas are also not neoplastic lesions. Many a times, they are found in the form of rests, without forming a mass lesion. Although neuromuscular hamartoma, composed of neural and skeletal muscle differentiation, has been referred to as "benign triton tumor," it is technically incorrect. This has also been proved by karyotyping.^[3] Some clinical details may aid in distinguishing the oral leiomyomatous hamartoma from other gingival lesions in newborns. The gingival cysts are round and small lesions and present white or cream color. Vascular malformations often have reddish surface and tend to decrease in size after compression. The melanotic neuroectodermal tumor of infancy is presented as a pigmented lesion, usually located in the anterior alveolar ridge of the maxilla, characterized by a rapid growth, different from the features showed by congenital epulis. Oral teratoma and rhabdomyosarcoma are rare tumors that arise as a solid mass with progressive and invasive growth.^[5] Although very rare, peripheral odontogenic tumors in newborns were also reported and should be included in the differential diagnosis.^[6] The congenital epulis cells are negative for neural, fibroblastic, myofibroblastic, myogenous, vascular, and histiocytic markers, and there are not evidences that congenital epulis grows after the birth.^[7] The majority of benign tumors in the young are probably developmental rather than true neoplasms. Differences found in various population group children and adolescents and those from other countries may be attributable to genetic and geographic differences.^[8] Treatment consists of simple conservative excision under general or local anesthesia; no recurrence has been reported. Surgery should not be radical as it minimizes danger of damaging underlying alveolar bone

Table 1: Summary of reported cases of leiomyomatous hamartomas of the midline maxillary gingiva^[9-12]

Authors	Year	Age	Sex	Location	Age of first presentation	Size (cm)	Clinical diagnosis
Takahashi <i>et al.</i>	1962	2.5 months	Female	Median maxilla	20 days	0.4×0.5×0.7	Congenital epulis
Mushimoto <i>et al.</i>	1982	11 months	Female	Median maxilla	At birth	0.5×0.5×0.7	Congenital epulis
Kajiyama <i>et al.</i>	1983	4 years 5 months	Female	Median maxilla	At 5 months	1.5×0.6×0.7	Congenital epulis
Kanekawa <i>et al.</i>	1989	3 years	Male	Median maxilla	At birth	0.5×0.5×0.5	Congenital epulis
Seki <i>et al.</i>	1991	2 years 3 months	Female	Median maxilla	At birth	0.8×0.5×0.4	Congenital epulis
Semba <i>et al.</i>	1993	2 years 2 months	Male	Median maxilla	At birth	0.5×0.5×0.4	Congenital epulis
Misawa <i>et al.</i>	1994	1 years 7 months	Female	Median mandible	At birth	0.3×0.2	Congenital epulis
Takeda <i>et al.</i>	2000	10 months	Male	Median maxilla	At birth	0.6×0.6×0.6	Congenital epulis
Lida <i>et al.</i>	2007	2 years 7 months	Male	Median maxilla	2 years 5 months	0.5×0.3×0.4	Benign tumor
Zhang <i>et al.</i>	2008	2 years	Female	Median maxilla	5 months	0.5×0.5×0.7	Congenital epulis
Alqahtani <i>et al.</i>	2013	18 months	Male	Labial median gingiva maxilla	18 months	3×2×3	Congenital epulis
Damm <i>et al.</i>	2014	-	-	Incisive papilla	-	-	Nodule of incisive papilla
Present case		2 years	Female	Incisive papilla	At birth	2.5×0.5×0.5	Incisive papilla growth

and developing tooth buds. Delay in operation may cause airway obstruction and feeding difficulty.

Conclusion

Oral Smooth muscle (leiomyomatous) hamartomas should be considered in the clinical differential diagnosis of congenital epulis. Early and prompt surgical excision is the widely preferred approach in this condition. It is important for dental professionals to be aware of this congenital tumor and its presentation, differential diagnosis, histopathology, and management. A multidisciplinary approach is required for managing such patients by considering a significant reduction in parent's anxiety and patient's morbidity.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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