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Maxillary Gingival Vascular Hamartoma: In A 8-Year-Old Female with a 10 Years Follow-Up

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1. Abstract

Oral hamartomas are considered as malformations of oral tissues and the usual ones are usually composed of lymphatic and blood vessels. The aetiology of hamartomas is unknown. Gingival hamartomas are most commonly found on the anterior maxillary or mandibular gingiva, but they can arise somewhere in the oral cavity. We report a gingival hamartoma in a child of 8 years old. Clinical examination revealed a lesion that was non-ulcerated, pedunculated and partially enveloped the two maxillary central incisors and the left lateral and the left erupted canine buccally and palatially. The questionary of her parents revealed no medical or family history. Panoramic x-ray doesn't show any bone lesion of the mentioned region. The preliminary diagnosis balanced between some tumour-like lesions as pyogenic granuloma, lobular capillary hemangioma, giant cell granuloma, or tumour lesions like epithelioid hemangioendothelioma, hemangiomas, angiosarcoma and hamartomas. MRI was proposed to avoid any vascular lesion or tumour. Appearance of limited hypertrophy with gingival thickening presenting a high signal in T2 without alteration of the cortical bone of the alveolar arch without any sign of malignancy. Under local analgesia, a complete surgical excision of the lesion followed by electro-coagulation. Specimen was sent for histological evaluation and the result was as a vascular hamartoma. Follow-up was carried out 15 days after, 1 months and one year later and no recurrence was observed. Ten years after the maxillary gingival contour witnessed the establishment of a normal gingival contour.

2. Introduction

The term hamartoma, from the Greek ἁμαρτία, hamartia ("error"), originally described by Albrecht in 1904 and was used to describe a "tumor- like" malformations or congenital errors of tissue development, looks like a focal overgrowth of regular cells, but principally non-neoplastic [1]. Hamartomas are usually found in some organs as pancreas, lungs, spleen, liver and kidneys and rarely observed in the head and neck region [2,3]. Oral hamartomas are considered as malformations of oral tissues and the usual ones are usually composed of lymphatic and blood vessels [4]. Nevertheless, some rare entities are composed of epithelial, fibrous, neural tissues [5] and infrequently smooth muscles are found [6]. The aetiology of hamartomas is unknown [3, 5-7]. They replicated a local malformation of a tissue owing to an overgrowth of cells from the ectoderm, different from neoplasm or tumour where the growth is originated from a single mutated cell (monoclonality), where the neoplastic cells are clonally originated [8]. They are rare, represent less than 1% of all oral lesions, are more frequently detected in children and young adults and can arise at several ages with no gender predilection [3]. Clinically they appear as small, pink or red, firm, painless masses on the gingiva. These lesions are often solitary and well-circumscribed, and they may be sessile or pedunculated. Gingival hamartomas are most commonly found on the anterior maxillary or mandibular gingiva, but they can arise somewhere in the oral cavity [3]. The diagnosis of gingival hamartomas is based on histological analysis of a biopsy specimen. Immunohistochemical staining can be useful in distinguishing gingival hamartomas from other oral lesions, such as fibromas, pyogenic granulomas, angiosarcoma [9-11]. The treatment of hamartoma is based on a surgical excision with a margin of normal tissue to prevent recurrence and relapse is barely observed [12]. We report a case of gingival hamartoma in a child of 8 years old with respect to CARE guidelines [13] with 10 years follow-up.

3. Case Report

A young 8-year-old female patient was referred to our clinic by her pedodontics' for the evaluation of an uncommon gingival tumefaction in relation of the upper central incisors in the maxilla. The lesion was asymptomatic with a red color. Her parents testified that it was present from at least one year and had gradually grown-up. Clinical examination revealed a good oral hygiene, plaque overgrowth was ruled out and the lesion was non-ulcerated, pedunculated and partially enveloped the two maxillary central incisors and the left lateral and the left erupted canine buccally and palatially (Figure 1a and 1b). The questionary of her parents revealed no medical or family history. Panoramic x-ray doesn't show any bone lesion of the mentioned region (Figure 2). The preliminary diagnosis balanced between some tumour-like lesions as pyogenic granuloma, lobular capillary hemangioma, giant cell granuloma, or tumour lesions like epithelioid hemangioendothelioma, hemangiomas, angiosarcoma and hamartomas. After discussion with the family, an MRI was proposed to avoid any vascular lesion or tumour. Examination was proceeded with a high field 1.5 T. Sequences used: spin-echo pondered in T1 in the axial plane of 5 mm, and in T2 before and after suppression of the fatty signal. Appearance of limited hypertrophy with gingival thickening presenting a high signal in T2 without alteration of the cortical bone of the alveolar arch without any sign of malignancy. (Figure 3). Under local analgesia, a complete surgical excision of the lesion followed by electro-coagulation. (Figure 4). Specimen was sent for histological evaluation and the result was as: a limited proliferation tumour located in the chorion. Stratified squamous epithelium hyperkeratosis with a corrugated surface and acanthosis. Tapered rete ridges, no dysplasia and vertically oriented irregular vascular section lined by a hyperplastic and irregular endothelium. Some vessels show intraluminal papillary projections and the diagnosis was as a vascular hamartoma and immunohistology was negative for CD34 to eliminate the diagnosis of angiosarcoma (Figure 5a, 5b and 5c). Follow-up was carried out 15 days after, 1 months and one year later (Figure 6a and 6b). No recurrence was observed and a complete healing with a complete eruption of the left canine were noted. Ten years after the maxillary gingival contour witnessed the establishment of a normal gingival contour. (Figure 7).

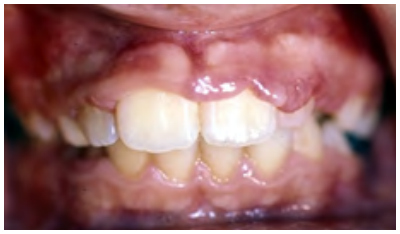

Figure 1a: Buccal view of the tumour.

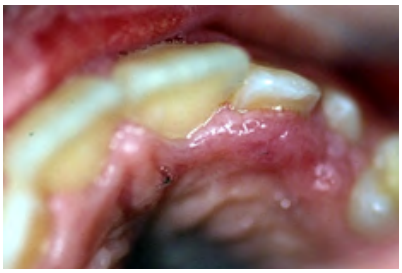

Figure 1b: Palatal extension of the tumour.

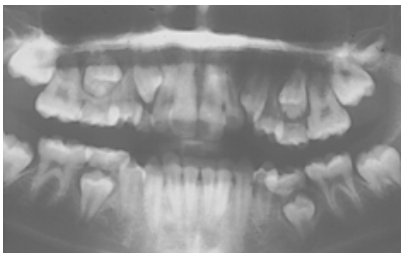

Figure 2: Panoramic x-ray that does not reveal any bony extension.

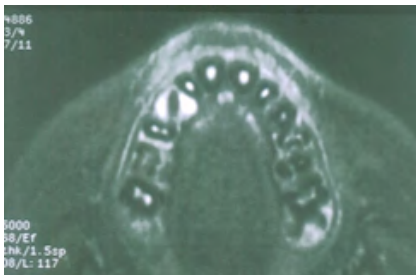

Figure 3: Appearance of limited hypertrophy with gingival thickening presenting a high signal in T2 without alteration of the cortical bone of the alveolar arch without any sign of malignancy.

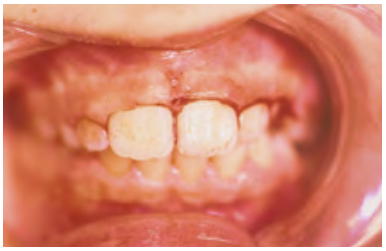

Figure 4: surgical excision of the lesion followed by electro-coagulation.

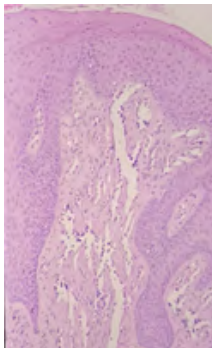

Figure 5a: Photomicrograph showing a limited proliferation tumour located in the chorion covered by a stratified squamous epithelium with a corrugated surface and acanthosis (→). Large irregularly shaped vessels with hyperplastic and irregular endothelial projection (→) are seen. (H&E stain x 50).

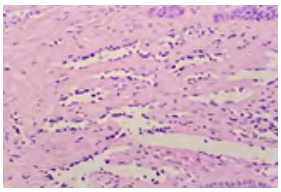

Figure 5b: Photomicrograph showing that some vessels with intraluminal papillary projections and the endothelial cells are inside the lumen of the vessels ( ). (H&E stain x 100).

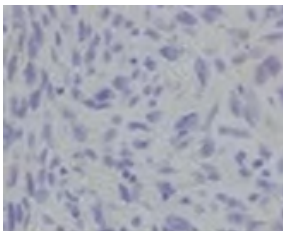

Figure 5c: Immunohistology was negative for CD34 that eliminate the diagnosis of angiosarcoma.

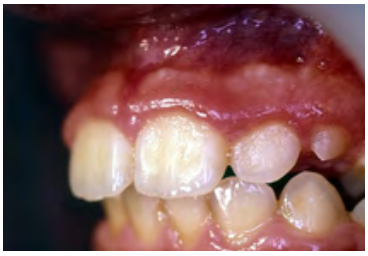

Figure 6a: buccal view, no recurrence was observed and a complete healing with a complete eruption of the left canine were noted at six months.

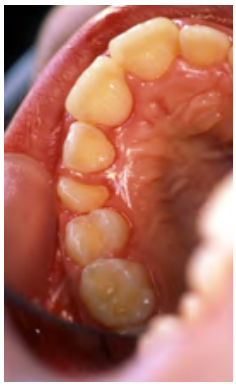

Figure 6b: palatal view a total remission of the palatal gum.

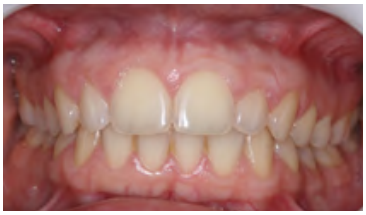

Figure 7: Ten years after the maxillary gingival contour witnessed the complete healing and the normal healthy of the gingival contour.

4. Discussion

Gingival hamartoma is very rare, native cells that influence in hamartomas developments including epithelial derivatives as nerve, muscle, bone, vessel, and fat cells from odontogenic and non-odontogenic origin [3, 5, 7, 12, 10,14]. The most frequent localization of hamartomas cases is reported in the tongue and palate followed by maxillary gingival region with a female predilection 1/ 3 and detected between 2 months to 19 years. The maxillary incisive papilla region is the most frequent in the maxilla and the size of the lesion is less than 1 cm [15, 16]. An English and Japanese literature search for oral gingival hamartoma until now yielded only 18 cases reported in the maxillary alveolar and gingival region. The remainder of the cases are reported in other areas like the tongue and palate. All the 18 cases including our case occurred in the median maxillary region. Further the reported cases were in the posterior part of the tongue, in the gingiva and hard palate were less stated [3,7,9,10]. (Table 1) [7, 15, 17-30]. Clinically appears as non-ulcerated sessile or pedunculated, uni or multilobular mass. [6] The recommended treatment is the simple excision and recurrences have not been reported. [5,6] They are asymptomatic and complications are moderate except when they are situated at the base of the tongue [14]. In the oral cavity, hemangiomas and lymphangiomas represents 30% and 60% of all benign vascular and soft-tissue tumors with 75% of lesions at birth and 85% after the first year of age and these tumours can make the diagnosis of hamartoma very difficult [31, 32]. The most common tumour in the oral and maxillofacial regions in infant and young children are hemangioma's and they reporting 11% of all kind of tumors and 38.5% of benign one [33, 34]. The follow-up period in the literature varies between 6 months and 5 years. In our reported case the follow-up period was over ten years. (Table 1). The malignant vascular tumors in the oral cavity of young children are very rare and represent less than 1% of vascular tumours [35]. Tumours that appear at childbirth or later on are stated as hereditary and thus as tissue abnormalities and not neoplasms [35]. In children few benign tumors of the head and neck necessitate magnetic resonance imaging (MRI) examination to analyse whether the surrounding tissues and organs are intricate and the degree of invasion [35, 36]. In this paper we reported a gingival tumour on the maxillary gingiva which was illustrated by an unusual endovascular papillary aggregate of endothelial cells with a final histological diagnosis as gingival vascular hamartoma. The lesion initially triggered a diagnostic and treatment dilemma. The surgical excision was the appropriate treatment choice.

Table 1: Clinical case report of gingival hamartomas in the literature.

	Author	Year	Age	Gender	Site	Treatment	Recurrence	Follow-Up
1	Takahashi et al.	1962	3 months	F	Anterior Median Maxilla	Excision	No	6 months
2	Mushimoto et al.	1982	11 months	F	Anterior Median Maxilla	Excision	No	9 months
3	Kajiyama et al	1983	4 years	F	Anterior Median Maxilla	Excision	No	6 months
4	Kanekwa et al.	1990	3 years	M	Anterior Median Maxilla	Excision	No	1 year
5	Seki et al.	1991	2 years	F	Anterior Median Maxilla	Excision	No	6 months
6	Ng et al. 1992	1992	3 months	F	Incisive Papilla	Excision	No	6 months
7	Semba et al.	1993	2 years	M	Anterior Median Maxilla	Excision	No	5 years
8	Blum et al.	1994	11 years	M	Mandibular Giniva	Excision	No	3 years
9	Misawa et al	1994	1 year	F	Anterior Median Maxilla	Excision	No	6 months
10	Nappier et al	1996	5 years	F	Hard Palate	Excision	No	1year
11	Takeda et al.	2000	10 months	M	Anterior Median Maxilla	Excision	No	1 year
12	Correa et al.	2001	6 years	F	Incisive Papilla	Excision	No	2 years
13	Kujan et al.	2005	11 months	M	Anterior Median Maxilla	Excision	No	1 year
14	Oliviera et al.	2005	10 months	M	Anterior Median Mandible	Excision	No	6 months
15	Scarpelli et al.	2007	6 months	F	Anterior Median Maxilla	Exc ision	No	7 months
16	Raghunath et al.	2016	15 years	F	Anterior Median Maxilla	Excision	No	0
17	Elo et al.	2017	34 years	F	Median Maxilla	Excision	No	0

5. Conclusion

Gingival vascular hamartoma is a rare benign lesion that arises from the gingival tissue. These lesions are typically asymptomatic and are often discovered incidentally during routine dental examinations. The diagnosis of gingival hamartomas is made through clinical examination and histological analysis of a biopsy specimen. Surgical excision is the treatment of choice for larger or symptomatic lesions, with an excellent prognosis after excision.

6. Consent Form

Patient's parents signed a written consent form.

7. Ethical Approval

None.

8. Disclaimers

The author does not have any financial interests, either directly or indirectly, in the products or information listed in this paper.

9. Competing Interests

None

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