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Oral Angioleiomyoma: A Rare Pathological Entity

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Abstract. *Leiomyomas are uncommon in the oral cavity and rare on gingiva. They account only for 0.42% of all soft tissue lesions in the oral cavity. We present an extremely rare case of leiomyoma localized to the attached gingival, simulating an epulis in a healthy 14-year-old boy. The tumour was described at the clinical and instrumental level; moreover, its histopathological phenotype was depicted. The treatment of the choice was the radical excision. The wound was closed by surgical dressing with 2-0 silk suture.The post-operative course was uneventful. The surgical wound healed in one week with normal scarring. Finally, the problems of differential diagnosis with other tumours of the oral cavity and the most appropriate therapeutic procedures are discussed.*

Leiomyomas (LMs) are benign, smooth muscle neoplasms that may develop from aberrant smooth muscle cells (SMCs), or their precursors in the media of blood vessels, in the muscularis of the gut and in the body of the uterus (1, 2). Although they can appear in any location, preferred sites are the uterus, the gastrointestinal tract, the lung and the skin (3, 4). LMs can be divided based on their localization into superficial LM when growing in subcutaneous tissue and in deep LM when they are found in the deep somatic soft tissue, retroperitoneal or oral/abdominal cavity (1, 4). According to the World Health Organization, LMs are classified into three histological groups: a) vascular (angioleiomyomas), 74% of cases; b) solid leiomyomas, 25%; c) epithelioid (leiomyoblastomas), fewer than 1% (1, 3-5).

Due to the paucity of smooth muscle, LM is rare in the oral cavity and there are fewer than 200 cases reported in the literature (5). These tumours account for only 0.42% of all soft tissue lesions reported in the oral cavity and only 0.065% of LMs have this location (4, 6). In the mouth, the lips are the most common site, followed by the tongue, cheeks, palate and, more rarely, gingiva and retromolar trigon (7, 8). Concerning the origins o SMCs in the oral cavity, vessel walls, the circumvallate papilla, and atypical arrectores pilorum muscle as adnexial SMCs in the cheeks are cited (9-11). Frequently in the oral cavity LM tends to occur along the midline (site of fusion and errors during embryological development, such as the nasopalatine foramen and tongue).

Epidemiologically, LMs can grow at any age with preponderance in adult patients, with gender predilection based on the reports of various authors (1, 4, 5). Clinically, LMs are unspecific masses, with several aspects from normal to more congestive mucosa, with the colour of the overlying mucosa being ischemic, normal, hyperaemic, or purple, according to vascularisation and site (1, 2, 4, 13-18). The size of LMs in the oral cavity can vary from a few centimetres to giant forms (leiomyomatous hamartoma of the tongue) (1, 3, 4, 13). Difficulty in deglutition, toothache, loose teeth, and pain have been reported, all signs probably due to local ischaemia and compression of nerves close to the tumour; these symptoms, usually, appears after several years of development (9). Microscopic differential diagnosis is required to distinguish LM from many benign tumours such as congenital gingival granular cells epulis (a nodular lesion of the newborn, negative for S-100 reaction with unknown origin), granular cell myoblastoma (a nodular lesion with granular cytoplasm of its cells derived from Schwann cells, with positivity for S-100 protein), fibromas, lipomas, salivary gland neoplasms, hemangiomas, cartilaginous choristoma of the tongue (in case of appearance on the midline of this site), thyroid ectopic nodules, non angiomatous hamartomas, an incisive papilla cyst or nasopalatine duct cyst, fibroepithelial polyps, intraoral rhabdomyomatous hamartoma, and, logically, the malignant counterpart of leiomyoma, *i.e.* leiomyosarcomas (2, 14, 15). The most frequent lesions considered in the differential diagnosis are fibroma, due to its innocuous evolution and clinical features, and fibrous epulis due to its site (11).

Case Report

In October 2008, a healthy Caucasian 14-year-old male was referred to the Department of Oral Surgery of the School of

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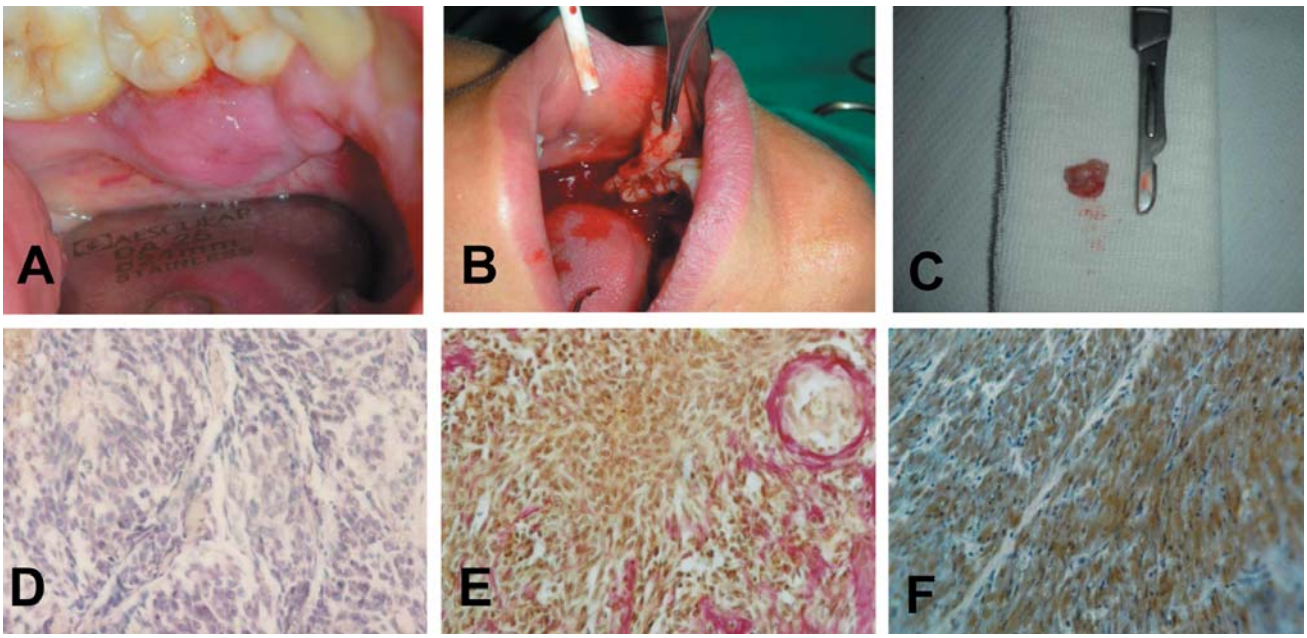

Figure 1. *A: The clinical appearance of the tumour. B: Intra-operative appearance of the tumour. C: Macroscopic appearance of the tumour. D: The histological pattern of the tumour revealed a well circumscribed nodule with prominent vascular spaces and redundant smooth muscle cells with a fascicular arrangement (haematoxylin-eosin staining; original magnification ×20). E: The tricromic staining of Van Gieson better demonstrates the vascular spaces within the neoplasm (haematoxylin-Van Gieson; original magnification ×20). F: Immunohistochemical staining for actin demonstrates the muscle origin of the neoplastic cells (ABC, original magnification ×20).*

Dentistry of the Second University of Naples, Italy, for evaluation and treatment of a small mass of the lingual mucosa of the left mandible in the premolar area. The patient had been aware of the lesion for 3-4 months, without experiencing any discomfort. The mass had increased in size slowly. Intraoral examination disclosed a nodular hemispherical, well-circumscribed mass of about 1-2 cm in diameter (Figure 1A), the overlying mucosa appeared normoemic, normotrophic, not bloody; lymph nodes were not involved. On palpation, the lesion was not painful, with fibroelastic consistency. Radiographs were negative for bone lesions.

The mass being considered as a tumour was classified as T1, N0, M0. The treatment of the choice was radical excision (Figure 1B) with 2-mm free margins; with the patient under local anaesthesia we removed the tumour easily. There was no relation between the tumour and underlying bone. The wound was closed by surgical dressing with 2-0 silk suture. Antibiotic coverage and chlorhexidine gluconate were used prophylactically. The post-operative course was uneventful. The surgical wound healed in one week with normal scarring.

The surgical specimen consisted of an encapsulated mass measuring just under 2 cm (Figure 1C). The excised biopsy tumour specimen was fixed in 10% buffered formalin and was paraffin-embedded. Five-μm-thick sections were stained with haematoxylin-eosin, haematoxylin-van Gieson and Periodic acid Schiff-haematoxylin. Microscopic examination showed a well-delimited subepithelial proliferation of small fusiform cells that formed bands, with no clear evidence of a capsule although well delimited. These fascicles were irregularly scattered, mixed up with other elements, such as lymphatic and venous vessels, and nerve fibres of variable size (Figure 1D). The predominant elements were SMCs. Histological stain with Van Gieson, as well as immunohistochemical staining (expression of muscle-specific actin) confirmed the muscle origin of the neoplastic cells, which were also negative for S100 (Figure 1E and 1F). During the one-year follow-up, no abnormalities were noticed. There has been no evidence of recurrent disease to date. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Discussion

The aetiology of LM is obscure. Chronic inflammation, continuous mechanical trauma and spontaneous development may be mentioned as the causes for the origin in the oral cavity. Nowadays, LM should be considered as a result of chronic irritation that can determinate the growth of a smooth muscle tumour in patients with genetic susceptibility. Congenital epulis and granular myoblastoma resemble the

clinical findings of our case but the first is not relevant considering the age of the our patient and the second exhibits a strong positivity for S-100 protein (16). The most important pathologies in differential diagnosis, especially in terms of the histological patterns, includes neurofibroma, which is identified by positivity of S-100 protein, and leiomyosarcoma, which can be diagnosed by counting the number of mitotic figures per field: a count of more than 5-10 reveal a malignant behaviour (1).

The treatment of choice consists in local radical excision with 2-mm lateral free-margins of the tumour. Simple enucleation may be insufficient to prevent recurrence. In the bone, the operation may consist of excision with surgical curettage with a clear bone border without sacrificing functional structures. By clinical and radiological follow-up it is possible assess for recurrence. The prognosis of true LM is very good. Metastasis of such tumours has not been reported. Local recurrence may be present when the excision is incomplete.

The final diagnosis, in our case, derived from histological examination (rare mitoses, less than 5 per field, absence of cellular atypia), histochemical (Van Gieson staining) and immunoistochemical positive patterns (expression of muscle specific actin and no expression of S100). Therefore, the surgical treatment was successful and patient did not experience recurrence.

Finally, it should be underlined that hamartomatous form of LMs have been reported in the oral cavity. They are composed of well-differentiated SMCs together with normal tissue in a site where they normally grow: *i.e.* abnormal tissue without normal histological architecture; clearly, the major component is the smooth muscle tissue (18). Multiple leiomyomatous hamartomata have been reported in the oral cavity (19).

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