ARTICLES SCIENTIFIQUES

Granular cell tumors of tongue

Tumeur à cellules granuleuses de la langue

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Abstract

Granular cell tumors (GCT) are ubiquitary tumors with a predilection for the head and neck region. At the oral level, they are often located on the tongue. They are more frequent in women with a sex ratio of 3/1 and a peak incidence in the fourth and fifth decades of life. They are most often benign. Malignant transformation has been reported in 1–3% of cases. They are characterized by a proliferation of large cells with abundant granular eosinophilic cytoplasm. Because of their potential recurrence and their morphology that are similar to malignant tumors, complete surgical excision is recommended. Immunohistochemical analysis often helps in diagnosis. We report the case of a 45-year-old woman consulting for a 1 cm nodule, evolving for 5 months. Surgical excision was performed. Histological examination showed the presence of a non-encapsulated proliferation infiltrating the striated muscle. The overall findings concluded a granular cell tumor without histological criteria of malignancy. The postoperative course was uneventful. At 18 months postoperatively, the patient had no recurrence.

Key words

abrikosoff's tumor, tongue, surgery, histology, Immunohistochemistry

Résumé

Les tumeurs à cellules granuleuses (TCG) sont des tumeurs ubiquitaires avec une prédilection pour la région de la tête et du cou. Au niveau buccal, elles sont souvent localisées sur la langue. Elles sont plus fréquentes chez les femmes avec un sex-ratio de 3/1 et un pic d'incidence dans les quatrième et cinquième décennies de la vie. Ils sont le plus souvent bénins. Une transformation maligne a été rapportée dans 1 à 3 % des cas. Ils se caractérisent par une prolifération de grandes cellules avec un cytoplasme éosinophile granuleux abondant. En raison de leur potentiel de récurrence et de leur morphologie similaire à celle des tumeurs malignes, l'excision chirurgicale complète est recommandée. L'analyse immunohistochimique aide souvent au diagnostic. Nous rapportons le cas d'une femme de 45 ans, qui a consulé pour un nodule de 1 cm évoluant depuis 5 mois. L'excision chirurgicale a été réalisée. L'examen histologique a montré la présence d'une prolifération non encapsulée infiltrant le muscle strié. En conclusion il s'agit d'une tumeur à cellules granuleuses sans critère histologique de malignité. L'évolution postopératoire s'est déroulée sans incident. À 18 mois la patiente n'avait pas de récidive.

Mots clés

tumeur d'abrikosoff, langue, chirurgie, histologie, immunohistochimie.

INTRODUCTION

First reported in 1926 by Abrikossoff, Granular Cell Tumor (GCT), also known as “Abrikossoff's tumor” or “granular cell myoblastoma”, is a benign rare tumor of soft tissue. It presents often as single, insidious in origin, painless, slow-growing, well-delineated lesion in the fourth to sixth decade of life. Characterized by the accumulation of plump cells with abundant granular cytoplasm. The etiology of this disease is controversial and much discussed in the literature. The initially suggested muscular origin has been dismissed in favor of a nervous origin, or more precisely a schwannian origin, that for a long time was considered to be of myoblastic origin, a few of the reports indicating association with a traumatic episode. Lesion may appear everywhere in the body, but over 50% of cases occur in the head and neck. In the oral cavity, the most affected site is the tongue although, other sites such as the hard palate, buccal mucosa, lip, uvula, parotid gland and gingiva have been reported. Mainly on the tongue. Its extra-tongue localization is rare. The diagnosis of certainty is exclusively histological and immunohistochemical. About 2% of cases exhibited

malignant features. Different treatment approaches from regular monitoring to radical excision have been suggested (1-5).

OBSERVATION

Patient aged 45, with no medical or surgical history, consulted the department of oral medicine and surgery at the university clinic of Monastir, for a nodule on the tongue that had been present for 5 months.

- Exobuccal examination revealed no abnormalities.
- Endobuccal examination revealed a single, sessile nodule in the middle third of the dorsal surface of the tongue, 1cm in diameter, with a firm consistency, covered by a depilated, erosive mucosa, bordered by supermented, splayed edges and bordered posteriorly by a depilated macule, 1.5 cm in diameter. "Figure1"

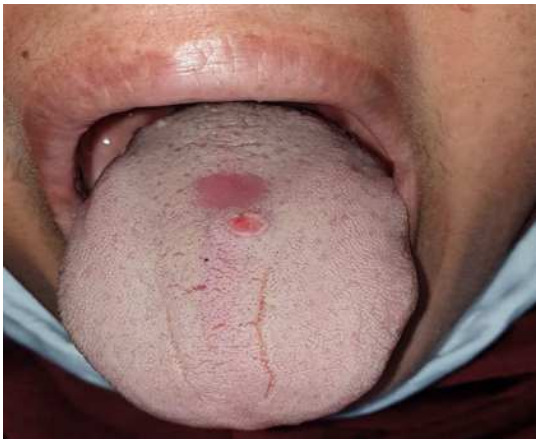

Figure 1 Sessile nodule in the middle third of the dorsal surface of the tongue, 1cm in diameter, with a firm consistency, covered by a depilated, erosive mucosa, bordered by supermented, splayed edges and bordered posteriorly by a depilated macule, 1.5 cm in diameter.

An exeresisal biopsy was taken, straddling healthy and pathological tissue. the operative specimen was sent for anatomopathological examination, which showed tumor proliferation in the chorion, with patches of large cells with abundant granular and eosinophilic cytoplasm and small, monotonous nuclei. Mitoses were absent. The tumor dissociated muscle cells. The diagnosis of granular cell tumor of the tongue was therefore retained. "Figure 2A, B" Checks are made after ten days, after 6 months and after 18 months which show no signs of recurrence. "Figure3"

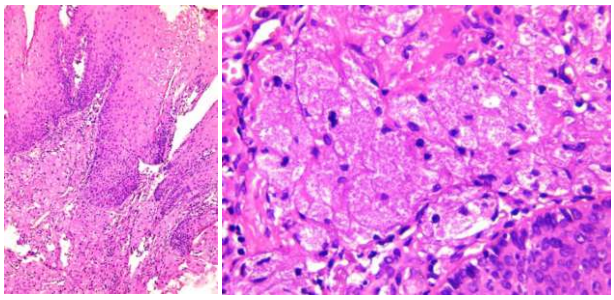

Figure 2 (A) Beneath a hyperplastic epidermis, the dermis contains a mesenchymal tumor made up of large cells (HE x 100); (B) The tumor cells have a small, round nucleus and abundant, eosinophilic, granular cytoplasm (HE x 400).

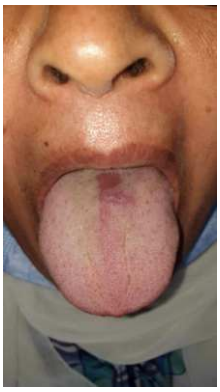

Figure 3 Control of lesion site after 18 months showing no sign of recurrence

DISCUSSION

Abrikossoff's tumor was first discovered in 1929 - 1926 by the Russian pathologist aq Alexei Ivanovich Abrikossoff. It is a rare benign non-epithelial tumor (6). The prevalence of Abrikossoff's tumor is only 0.019-0.03% of all tumors. Lesion may appear everywhere in the body, subcutaneous tissue, larynx, trachea, bladder, uterus, orbit, parotid gland, vulva, and central nervous system, but over 50% of cases occur in the head and neck, and particularly the oral cavity (50-70%). Lack and al suggested that the frequency of TCG in the oral cavity may be favored by repeated trauma from the teeth. In the oral cavity, the tongue represents the main location (23-28% of cases) followed by the floor of the mouth and the palate. On the tongue, 48% of TCGs occur on the dorsal surface, 15% on the lateral edges (as in this case) and the rest on the ventral side. The localization on the posterolateral of the tongue may suggest squamous cell carcinoma, especially if there is ulceration on the surface. Its extra-tongue localization is rare. As per the last large-scale systematic review in 2018, there were 1499 cases from 47 studies and 10.47% of all cases occurred in the tongue. Women are twice as affected as men which was explained by the frequent association with cancer of the esophageal or ENT cancer. GCT can develop at any age with a

revalence peak between the fourth and sixth decades of life. pediatric and congenital pediatric forms are exceptional. Darker pigmented individuals are more often affected than lighter pigmented individuals. A familial accumulation has been reported. (2,4,5,6,7,8).

Usually presents as a solitary asymptomatic nodule with a smooth or verrucous surface and fortuitously discovered. These are papules or nodules less than 3 cm in diameter, affecting subcutaneous or submucosal tissue, slow-growing, solitary, firm, mobile, whitish, painless, polyploid or sessile, granular, warty or ulcerated, and varying in color (pinkish, grayish or yellowish), they are slow growing (0.5 to 1 mm per year). In 12.46% of cases, there is a history of trauma or chronic inflammation. Lingual lesions are characterized by depapillation and atrophy of the overlying mucosa. The lesion is usually solitary, but cases of multicentric GCT have been reported. Clinical signs of malignancy include tumor size (> 40 mm), ulceration, rapid growth, invasion of adjacent structures and lymph node enlargement. The association of TCG with squamous cell carcinoma of the upper aerodigestive tract is not exceptional, accounting for 10% of cases. (2,3,4,7)

Histologically, the tumor consists of globular, polygonal or spindle-shaped cells arranged in clusters polygonal or spindle-shaped cells, arranged in clusters or trabeculae. The nuclei are small, hyperchromatic and often centrally located. Their cytoplasm is abundant, remarkable by the presence of eosinophilic granulations strongly stained by PAS; in electron microscopy, they correspond to grains of lysozyme at different stages of degradation. Approximately 50% of intraoral GCT lesions present pseudoepitheliomatous hyperplasia (PEH) associated with mucosal epithelium. If the biopsy is superficial, this appearance may raise suspicion of squamous cell carcinoma. This pseudo-epitheliomatous hyperplasia, owing often to epithelial reaction, often masquerades as glossal squamous cell carcinoma leading to mis-diagnosis. The histogenesis of GCTs remains unclear. Previously, GCTs have been considered to originate from skeletal muscles, neurons, fibroblasts, histiocytes, and myoepithelial cells. Moreover, degenerative changes or trauma-induced proliferation, which occur in a variety of normal and neoplastic cells, were also considered to be involved in the morphogenesis of GCTs. Recent reports based on immunohistochemical and electron microscopic studies of humans and animals have suggested that GCTs are derived from neural crest tissue, especially Schwann cells because of positivity for S-100 and vimentin. The cytoplasmic granules found in GCT cells have been suggested to be lysosomes, autophagosomes, or autophagolysosomes based on their ultrastructural features. Another common immunohistochemical marker that has been traditionally used for identifying GCT is CD68, a marker of lysosomes, mostly associated with macrophages, which is also usually positive in GCT, the cells also stain positively with Sudan black B and trichrome preparations. (1,4,5,6,7,8)

Clinically, the lesions resemble other neoplasms such as fibromas, lipomas, neurofibromas, neuromas, schwannomas, tumors of the small salivary glands and lymphomas. Also, the CGT has to be differentiated histologically from dermatofibroma, Reed neuroid spindle cell tumor, xanthoma and metastatic breast carcinoma, all of which may show granular cytoplasm. Diagnostic difficulty arises in cases where PEH is so pronounced that it can be mistaken for a squamous cell carcinoma (SCC), especially when superficial specimens of biopsies are collected, the majority of granular cell tumors are benign; malignant forms represent less than 2% of cases. The distinction between benign and malignant tumors is often difficult and is based on a combination of clinical and histological arguments. A size greater than 4 cm or rapid growth, the presence of rapid growth, the presence of necrotic and/or malignant areas (3,4,5,6,8,9).

The treatment of choice for single or multiple lesions of TCG, remains surgical excision with tumor-free margins, because the lesion is not encapsulate, often poorly limited, and it infiltrates the underlying muscle fibersas and the recurrence rate for GCTs with tumor positive margins is 21–50%. However, in the oral cavity, tissue preservation is essential, micrographic surgery may be indicated. Laser excision is recommended for small tumors. The radiotherapy and chemotherapy are not recommended for benign forms, because of the resistance of the tumor and the carcinogenic effect of these treatments. However, they are indicated for malignant forms. Spontaneous regression has been reported in exceptional cases. Malignant forms of TCG have a higher rate of recurrence rate (31-41%) with distant metastases in 50-63% of cases. Thus, a wide excision with margins of 2 to 3 cm is recommended by some authors with regional lymph node dissection. (4,5,6,7,8)

CONCLUSION

Oral GCT is rare it's an unifocal lesion, benign in the majority of cases. The diagnosis is anatomo-pathological and immunohistochemical. Oral localizations are relatively frequent. The exact role of trauma and inflammation in etiopathogenesis of GCT warrants further investigation. The dentist must include this entity in the list of in the list of differential diagnoses to be considered. The prognosis is favorable after simple surgical removal. In case of a positive diagnosis, a malignant form as well as an association as well as an association with a squamous cell carcinoma, although rare, must be eliminated.

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