CASE REPORT

Abrikossoff’s tumor of tongue: Report of an uncommon lesion

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ABSTRACT

Granular cell tumor, an uncommon neoplasm, can occur at any site in the body, but they are more commonly observed in the head and neck region, especially the oral cavity. A lesion of controversial origin, this entity has to be considered in the differential diagnosis of swellings of the tongue. Immunohistochemical findings obtained in the present case also render support for the neural histogenetic origin for this tumor. Though commonly observed in the fourth to sixth decades of life, we present this case in a 9-year-old female patient.

Key words: Abrikossoff’s tumor, granular cell myoblastoma, tongue

INTRODUCTION

Granular cell tumor also known as Abrikossoff’s tumor or granular cell myoblastoma was variably considered as being a true neoplasm, degenerative metabolic process or trauma-induced lesion. The most common site in the oral cavity is the tongue. It is especially seen in females in the fourth to sixth decades of life.

CASE REPORT

A 9-year-old female patient presented to the dental clinic with a chief complaint of a swelling on the left side of her tongue since 6 months. The swelling was initially small and a gradual increase in size was observed. There was no history of regression in size. Her past family, medical and dental history was not significant. On examination there was a solitary well-defined, non-tender and non-fluctuant swelling with smooth borders, on the left side of the dorsal surface of the tongue, measuring approximately 1.5 × 1 cm. Her hematogram findings were within normal limits. With a clinical diagnosis of a benign mesenchymal lesion, the lesion was excised under local anesthesia and the specimen subjected for histopathological examination. The hematoxylin and eosin-stained sections revealed a well-defined lesion surrounded by stratified squamous epithelium with the underlying connective tissue comprising of bundles of collagen and abundant muscle fibers. The deeper aspect of the connective tissue showed cellular proliferation of polygonal cells with abundant granular cytoplasm. The nucleus was located in the center and the cell membrane was indistinct. No mitotic figures were observed. A diagnosis of granular cell tumor was rendered. A panel of markers, namely, S-100, vimentin, cluster of differentiation (CD) 68, CD34, smooth muscle actin (SMA) and inhibin were used. Positivity was observed with vimentin, S-100, CD68 and inhibin [Figures 2, 3 and 4] confirming the diagnosis of granular cell tumor.

DISCUSSION

The granular cell tumor is also known as “Abrikossoff’s tumor, Abrikossoff’s myoblastoma, granular cell neurofibroma or granular cell schwannoma.” First described in 1926 by Russian pathologist Alexei Ivanovich Abrikossoff, it is an uncommon neoplasm. It was earlier also known as granular cell myoblastoma implying its origin from the striated muscles. A tumor of uncertain histogenesis, Abrikossoff’s tumor was considered to be a true neoplasm, degenerative metabolic process or a proliferative lesion induced by trauma. Earlier literature also suggested the other possibilities of muscular, histiocytic, fibroblastic or pericytic origin. With the advent of immunohistochemistry and the application of various markers, the hypothesis of neural origin has been widely accepted. The common location in the head and neck region is the tongue as was observed in the present case, followed by cheek mucosa palate, lip, gingiva, uvula and parotid gland. Other body sites being the skin, soft tissues, breast, lungs, nervous system, gastrointestinal tract, urinary bladder, female reproductive tract and bronchus. Typically appearing as a
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single (cases of multiple lesions have been reported) and sessile asymptomatic nodule, these lesions follow a benign course and the surface is smooth or pseudoulcerated. This ulceration, rapidity of growth and size can be confused with carcinoma. There are reports of painful symptoms during tooth brushing, consumption of spicy foods and bite trauma. Normally the size of the lesion is usually less than 3 cm, commonly affecting females in the fourth to sixth decades of life and is rare in children. Histopathologically these lesions present with round or polygonal cells with abundant granular cytoplasm and small nucleus that is eccentrically placed. The cells are arranged in the form of unencapsulated sheets, cords or nests with a syncitial appearance. Over 50% of cases, especially the lesions on the tongue show pseudoepitheliomatous hyperplasia. It was absent in the present case. The granularity of the cells has been attributed to various factors – neoplasia, degenerative or reactive processes like anoxia, metabolic disorders and lysosomic effects.

The differential diagnosis for this lesion includes other benign connective tissue tumors, traumatic fibromas, lipomas, neuromas, neurofibromas or schwannomas, with its malignant variants and even oral carcinoma, minor salivary gland tumors, dermoid cysts and vascular lesions. Granular cells can be found in lesions other than granular cell tumor, these include ameloblastoma, ameloblastic fibroma, odontogenic fibroma, odontogenic cysts, congenital epulis of the newborn and oral lichen planus. Ultrastructure and immunohistochemical findings have been a matter of research in these lesions. Additionally, it is clearly known that when there is damage to the myelin sheath, the myelin enters into a process of disintegration and is phagocytosed initially by Schwann cells and later by macrophages. Electronic microscopy has shown a continuous basal layer around the tumoral cells with certain resemblance to the perineurium and the presence of structures compatible with myelin inside the liposomes.

Immunohistochemistry has played a major role in confirming the neural origin of these tumors. Vimentin is a primordial member of the intermediate filament family and is expressed in mesenchymal tissue. S-100 immunoreactivity...
of granular cell tumors is the most widely used marker for identification of the granular cells, which constantly shows a strong and diffuse staining. It is a protein that belongs to a family of small acidic EF-hand (helix-loop-helix type conformation) calcium-binding proteins initially discovered in brain extracts. CD68 is a heavily glycosylated membrane protein, closely related to the family of lysosomal-associated mucin-like membrane proteins. It is positive in most of the cases of granular cell tumors. Inhibin-α is a glycoprotein hormone that participates in the regulation of the pituitary-gonadal feedback system. It is a specific marker for sex cord-stromal tumors of ovary and is also expressed in adrenal cortical neoplasms. It has been found to be positive in granular cell tumor. The therapeutic management of granular cell tumor is by conservative surgical excision.

CONCLUSION

Benign mesenchymal neoplasm usually presents as a swelling on the tongue. This lesion though uncommon should be considered in the differential diagnosis of swellings of the oral cavity and an updated knowledge of immunohistochemistry aids in the differential diagnosis of other lesions. Considering the aggressiveness of the granular cell lesions, a regular follow up of the patients is of pivotal importance.

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