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PRAKAZ SLUČAJA
CASE REPORT

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Histokemijska i imunohistokemijska procjena Abrikossoffova tumora jezika: prikaz slučaja

Histochemistry and Immunohistochemistry Evaluation of the Abrikossoff's Tumour of the Tongue: a Case Report

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Sažetak
Abrikossoffov tumor rijetka je dobroćudna neoplazma mekih tkiva i može se pojaviti na bilo kojem dijelu tijela, uključujući i orofacijalno područje. Obično je benigan, ali postoje podatci da je lokalno bio agresivan i maligan s udaljenim metastazama. Etiologija je nepoznata jer je u nekoliko istraživanja dokazano da zahvaća različite stanice. U prikazanom slučaju riječ je o Dominikanki u dobi od 36 godina koja je upućena na Odjel oralne i maksilofacijalne kirurgije poliklinike Federico Drugi u Napulju zbog ograničene i na opip čvoraste lezije od 17 milimetara na dorzumu jezika. Terapija se sastojala od ekscizijske biopsije na temelju dijagnostičke hipoteze o Abrikossoffovu tumoru, što je potvrđeno histopatološkom analizom te histokemijskom i imunokemijskom procjenom. Abrikossoffov tumor rijetka je neoplazma koja se mora oprezno dijagnosticirati nakon što se uzmu u obzir svi histološki i klinički parametri radi pravilnoga odabira terapije.

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Ključne riječi
tumor granularnih stanica; tumori mekih tkiva; jezik

Uvod

Abrikossoffov tumor (AT) rijetka je neoplazma mekih tkiva nepoznate etiologije (1), a prvi ga je opisao Abrikossoff liječeći pacijenta s lezijom na jeziku (2). Može se nazvati i gigantocelularni tumor (GCT) ili mioblastom (My), jer naziv te novotvorine ovisi o histogenezi koja nije točno određena, pa su zato različiti istraživači postavili mnoge hipoteze o njegovu nastanku, uključujući fibroblaste, mioblaste, nediferencirane mazenhimne stanice, Schwanove stanice, histiocite i neuralne stanice (3, 4).

Gigantocelularni tumor može zahvatiti bilo koji organ ili dio tijela. Većinom nastaje na glavi i vratu, posebice na jeziku, obraznoj sluznici i nepcu (5).

Može se pojaviti u svakoj dobi, no češći je između četvrtoga i šestog desetljeća, a rjeđe nastaje kod djece, premda u literaturi ima opisanih čak i takvih slučajeva (6). Od Abrikossoffova tumora dva ili tri puta češće obolijevaju žene negoli muškarci, a crna rasa pogođenija je od bijele (6, 7).

Introduction

Abrikossoff's tumour (AT) is a rare benign soft tissue neoplasm of unknown aetiology (1), first described in a patient with a lesion on the tongue by Abrikossoff (2).

It could be also called Granular cell tumour (GCT) or Myoblastoma (My), in fact, the denomination of this tumour depends on its real histogenesis, which remains unsettled, since different derivations have been postulated by various authors, including fibroblasts, myoblasts, undifferentiated mesenchymal cells, Schwann cells, histiocytes and neural cells (3,4).

Granular cell tumours can affect any organ or region of the body. Most GCTs occur in the head and neck region, especially in the tongue, cheek mucosa, and palate (5).

It can occur in patients of any age, although it is more common between the fourth and the sixth decades of life, being rare in children, although cases have been described in the literature even at a young age (6). Abrikossoff's tumour

Većina nedavno opisanih slučajeva nalazila se na bočnoj strani dorzuma jezika (8). Svrha ovoga rada bila je izvijestiti o gigantocelularnom tumoru na dorzumu jezika kod 36-godišnje pacijentice i kratak prikaz recentne literature o toj temi.

Prikaz slučaja

Dominikanka u dobi od 36 godina upućena je na Odjel maksilofacijalne kirurgije poliklinike Federico Drugi u Napulju s bezbolnom oteklinom na jeziku, slučajno otkrivenom prije pet mjeseci. Nije se žalila na krvarenje i nije bilo klinički važnih podataka u njezinoj povijesti bolesti (dijabetes, hipertenzija, alergije). Bila je zdrava i živjela je zdravo. Laboratorijski nalazi bili su uglavnom uredni. Klinički nalaz upućivao je na dobro ograničenu čvorastu leziju promjera 17 milimetara. Masa se nalazila ispod sluznice na bočnoj strani dorzuma jezika i imala je fibroelastičnu konzistenciju neosjetljivu na palpaciju. Sluznica iznad lezije bila je intaktna, bez ulkusa (slika 1.). Obavljena je ekcizijska biopsija na temelju diferencijalne dijagnoze traumatskoga fibroma jezika, lipoma i Abrikossoffova tumora (slika 2.). Histopatološki nalaz otkrio je da se novotvorina sastojala od velikih stanica s granuliranom citoplazmom. Lezija je zahvaćala cijeli dermis s istaknutim pseudoinfiltrativnim svojstvom i razdvajala je mišićna vlakna, što je upozorilo na tumor granularnih stanica. Tumor je bio ograničen unutar granica ekscidiranog uzorka i udaljen od dubokih struktura minimalno 0,3 milimetra (slis two or three times more common in women than in men. Black patients are more prevalent than white (6, 7).

Recently, most of the lingual lesions have been reported to occur on the lateral border of the dorsum of the tongue (8). Therefore, the purpose of this paper was to report a case of granular cell tumour of the tongue, in a 36-year-old female patient, which occurred on the dorsum of the tongue, together with a brief review of recent literature.

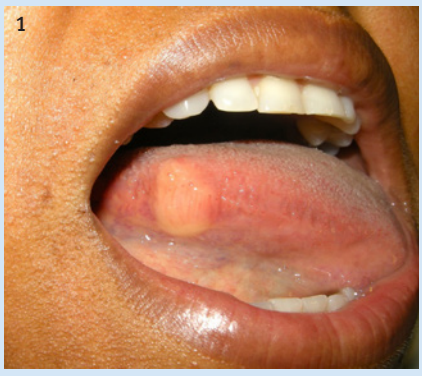
Case Report

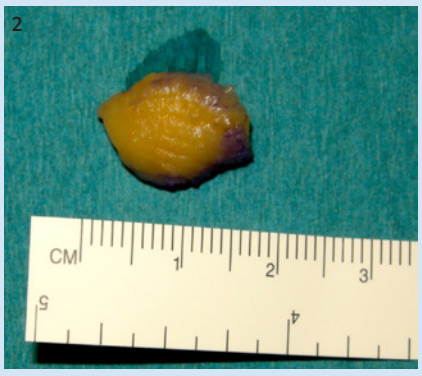
A 36-year-old Dominican was referred to the Department of Oral and Maxillofacial Surgery in Policlinico Federico II, Naples, Italy with a painless lingual swelling, incidentally discovered five months earlier. She did not complain of bleeding, and no significant clinical data (diabetes, hypertension, allergies) were present in her clinical history; our patient had always been well and she referred to a healthy lifestyle; laboratory investigations were mostly normal.

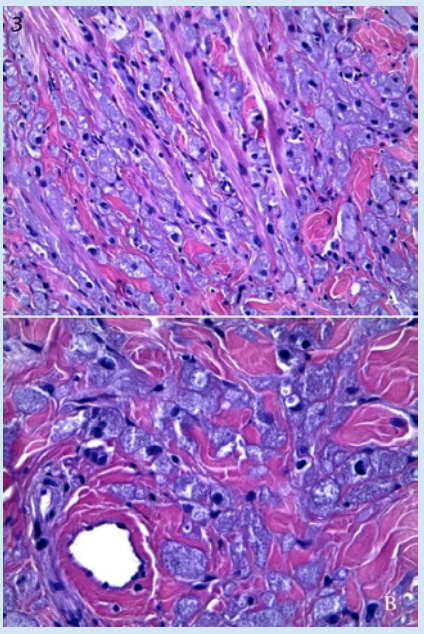
The clinical examination showed a well circumscribed lesion and sessile nodule, 17 mm in diameter. The mass was located just beneath the mucosa of the lateral border of the dorsum of the tongue, and had a fibroelastic consistency which was not tender to palpation. The overlying non-ulcerated mucosa was intact (Figure 1).

An excisional biopsy was performed based on a differential diagnosis of a traumatic fibroma of the tongue, lipoma, and Abrikossoff's tumour (Figure 2).

Histopathological findings revealed a neoplasia constituted of large cells with highly granular cytoplasm. The lesion involved entirely the dermis showing a pseudo-infiltra-







Slika 1. Dobro ograničena čvorasta lezija na dorzalnoj strani jezika
Figure 1 Well delimited nodular lesion located on the dorsum of the tongue
Slika 2. Veličina uklonjene lezije iznosi 1,7 cm preko najduže osovine uzorka
Figure 2 Excised lesion measuring about 1.7 cm across its major diameter.
Slika 3. Lezija zahvaća cijeli dermis, ima pseudoinfiltrativni oblik i razdvaja mišićna vlakna (hematoxilyn-eosin, A : 200 X i B : 250 X)
Figure 3 The lesion involves entirely the dermis and shows a pseudo-infiltrative pattern, dissecting the muscle fibres. (A, B: hematoxilyn-eosin, 200X and 250X, respectively).

ka 3.). Postoperativni tijek bio je uredan, pacijentica je nakon tjedan dana došla na kontrolni pregled, ponovno poslije tri, šest te dvanaest mjeseci i nije bilo znakova ponovne pojave neoplazme.

Rasprava

U slučaju lezije jezika posebnu pozornost valja posvetiti visokoj vaskularizaciji toga organa jer se tumorske stanice mogu brzo prenijeti cirkulacijom. Svakom se slučaju uvijek mora pristupiti diferencijalno dijagnostički jer kod naše pacijentice nismo bili sigurni je li riječ o Abrikossoffovu tumoru. Svojstva poput konzistencije i boje te palpacija rubova lezije mogu olakšati postavljanje dijagnostičke hipoteze (5, 9).

Histološki GCT obilježava proliferacija velikih poligonalnih neoplastičnih stanica sa zrncima i eozinofilnom citoplazmom, malim i ekscentrično smještenim jezgrama te nejasnim citoplazmatskim granicama (10). U nekim slučajevima epitel iznad tumora pokazuje znakove pseudoepitelomatozne hiperplazije (11). Histokemija i imunokemija mogu potvrditi dijagnozu jer se u nalazu vide S 100 pozitivne stanice s PAS-pozitivnim i CD 68-reaktivnim zrncima.

Dodatni imunohistokemijski prognostički čimbenici GCT-a uključuju Ki 67-indeks>10 % i p53 imunoreaktivnost, što u prikazanom slučaju nismo našli kao histološke kriterije malignosti (4, 12). Premda je Abrikossoffov tumor rijetka dobroćudna neoplazma, opisani su slučajevi malignih promjena GCT-a, uključujući pacijente kod kojih je nađeno više histoloških tipova malignosti. Zbog zloćudnoga potencijala ovaj tumor treba temeljito histopatološki analizirati. Podatci o veličini novotvorine, simptomima, brzoj progresiji, prodoru u okolne strukture i o regionalnim ili udaljenim metastazama vrlo su važni za histopatološku dijagnozu o dobroćudnosti ili zloćudnosti GCT-a (9). Terapija izbora za Abrikossoffov tumor je kirurška ekscizija sa sigurnim granicama rubova, premda to nije uvijek moguće jer novotvorina nema kapsulu nego je histološki nejasnih rubova (13). U prikazanom slučaju obavljena je ekscizijska biopsija. Angiero i suradnici opisali su neke slučajeve tumora GCT-a tretirane laserskom terapijom, ali dok su bili manji (14).

Zaključno, povezanost histološkoga i kliničkog nalaza s imunoprofilom temelj je za postavljanje točne dijagnoze u slučaju ove lezije.

Nakon kirurške ekscizije, samo daljnji opisi GCT-a te redovite kontrole pomoći će shvatiti i predvidjeti prirodu ovog tumora.

Izjava o sukobu interesa

Autori ističu da nisu u sukobu interesa.

tive pattern, dissecting the muscle fibres. For this reason, it was consistent with a granular cell tumour. The lesion was contained within the limits of excision with a minimum distance of about 0.3 mm from the deep structures (Figure 3).

The postoperative course was uneventful. The patient was first examined one week later and then, respectively, 3, 6, and 12 months after the surgical excision; so far, no sign of recurrence has been noted.

Discussion

In cases of tongue lesions, it is necessary to take utmost care since the high vascularization of this organ can lead to a rapid transportation of circulating tumour cells. In particular, we must always make a differential diagnosis, as in our case where the diagnosis of Abrikossoff's tumour was not certain. Features such as consistency, colour and the possible definition of lesion margins upon palpation may facilitate the establishment of diagnostic hypotheses (5,9).

Histologically, GCTs are characterized by the proliferation of large polygonal neoplastic cells with cytoplasmic granules, eosinophilic cytoplasm, a small and eccentrically located nucleus, and undefined cytoplasmic limits (10). In some cases, the epithelium that covers the tumour exhibits pseudoepitheliomatous hyperplasia (11). Histochemistry and immunohistochemistry can confirm the diagnosis of GCT when S100-positive cells containing pAS-positive and Cd68-reactive granules are seen. Adverse immunohistochemical prognostic factors of GCTs include Ki67-index > 10% and p53 immunoreactivity, in the present case we did not find any histological criteria of malignancy (4,12).

Although Abrikossoff's tumour is an uncommon benign neoplasm, cases of malignant GCT have been reported in the literature, including patients with more than one histological type of malignant GCT. In view of this malignant potential, the tumour should be submitted to thorough histopathological analysis. Data regarding the tumour size, symptoms, rapid progression, invasion of adjacent structures, and the presence of regional and distant metastases are of fundamental importance for the histopathological diagnosis of benign or malignant GCT(9).

Surgical excision with a safety margin is the treatment of choice for Abrikossoff's tumour, although this is not always possible because the tumour lacks a capsule, a condition histologically demonstrated by an undefined cell margin (13). An excisional biopsy was also the treatment of choice in the present case. Angiero et al. had also described some GCT tumour cases treated with laser but only when it is small in size (14).

In conclusion, the association of histological and clinical aspects with the immunoprofile is essential to establishing the correct diagnosis of this lesion.

After surgical excision, only further reports of the GCT along with the regular follow up reports will help us in understanding and predicting the nature of this tumour.

Conflict of interest

Authors have no conflict of interest to declare.

Abstract
Abrikossoff's tumour is a rare benign soft tissue neoplasm that can occur in any part of the body, including the orofacial region. The tumour is usually benign, but there are reports of cases in which the tumour shows a locally aggressive behaviour, malignancy, and distant metastases. The aetiology is unknown, since several studies have shown that different cells are involved. In the present case, a 36-year-old Dominican woman was referred to the Department of Oral and Maxillofacial Surgery in Policlinico Federico II, Naples with a circumscribed lesion and sessile nodule on the dorsum of the tongue measuring about 17 mm in diameter. The treatment consisted of an excisional biopsy performed on the basis of the diagnostic hypothesis of Abrikossoff's tumour, which was confirmed by histopathological analysis and histochemistry and immunohistochemistry evaluation. Abrikossoff's tumour is an uncommon neoplasm which must be carefully diagnosed considering all the histological and clinical aspects in order to be treated correctly.

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Key words
Granular Cell Tumor; Soft Tissue Neoplasms; Tongue

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