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CASE REPORT
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BENIGNI MEŠOVITI TUMOR MALE PLJUVAČNE ŽLEZDE

BENIGN MIXED TUMOUR OF THE MINOR SALIVARY GLAND

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Sažetak

Uvod: *Najčešća velika neoplazma pljuvačne žlezde je pleomorfni adenom. Samo 10% pleomorfnih adenoma javlja se u malim pljuvačnim žlezdama. U 42,8% do 68,8% slučajeva nepce predstavlja intraoralnu lokalizaciju ovog tumora Ovde je predstavljen redak slučaj pleomorfnog adenoma koji je ličio na leziju odontogenog porekla.*
Prikazu slučaja *je pacijent koji se javlja sa glavnim tegobama u vidu otoka u predelu gornjeg levog molara koji je trajao 3 meseca pre javljanja lekaru. Na ortopan snimku bila je dijagnostikovana promena mekog tkiva u levoj zadnjoj maksilarnoj regiji sa destrukcijom i resorpcijom alveolarnog nastavka. Razmatrana je radiografska diferencijalna dijagnoza benignih odontogenih/neodontogenih cista ili tumora, benignih malih tumora pljuvačne žlezde.Urađena je eksciziona biopsija i lezija je uklonjena u potpunosti do periosta poslata na histopatološki pregled, koji je potvrdio dijagnozu pleomorfnog adenoma.*
Zaključak: *Pleomorfni adenom je benigna neoplazma koja ima odličnu prognozu sa stopom izlečenja većom od 95%. Međutim, periodično dugotrajno praćenje je obavezno..*

Ključne reči: *Pleomorfni adenom, tumor pljuvačne žlezde*

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Abstract

Introduction: *The most common major salivary gland neoplasm is the Pleomorphic adenoma. Only 10% of the pleomorphic adenoma occur in minor salivary glands. 42.8% to 68.8% of cases with intraoral lesion occurs in the palate. Here we present a rare case of a pleomorphic adenoma masquerading as a lesion of odontogenic origin.*
Case report *presents a male patient who reported with a chief complaint of swelling in the upper left tooth region since 3 months. Panoramic radiograph revealed shadow of a soft tissue swelling in the left maxillary posterior region with destruction and resorption of the alveolar process. Radiographic differential diagnosis of benign odontogenic/non-odontogenic cysts or tumours, benign minor salivary gland tumours were considered. Excisional biopsy was carried out and the lesion was removed in total up to the periosteum and the overlying mucosa and sent for histopathologic examination, which suggested pleomorphic adenoma.*
Conclusion: *Pleomorphic adenoma is a benign neoplasm which has excellent prognosis with a cure rate of more than 95%. However, periodic long term follow up is mandatory.*

Key words: *pleomorphic adenoma, salivary gland tumour*

Uvod

Pleomorfni adenom je benigna neoplazma, koja je u većini slučajeva tipično neprimetna, sporastuća masa čvrste konzistencije. Malo je češća njegova pojava kod osoba ženskog pola. U većini slučajeva javlja se u velikim pljuvačnim žlezdama; međutim, nepce je takođe uobičajeno mesto za pojavu manjih pleomorfnih adenoma pljuvačnih žlezda[1].

Prikaz slučaja

Na Odeljenje oralne medicine i radiologije javio se pacijent sa glavnom tegobom u vidu otoka u predelu gornjeg levog poslednjeg zuba, prisutnog u poslednja tri meseca. Anamneza je otkrila da je pacijent uočio veoma mali otok u tom predelu pre tri meseca, koji je postepeno narastao do sadašnje veličine. Dva meseca pre toga, odnosno pre pet meseci, posetio je stomatologa radi vađenja gornjeg levog zadnjeg zuba. Nepuna dva meseca nakon ekstrakcije, pacijent je počeo da primećuje mali otok u pomenutoj regiji. Ovaj sporastući otok bio je praćen tupim bolom , bez većih promena u intenzitetu i nije zračio u susedne regije. Dve nedelje po pojavi otoka, pacijent je primetio mali čir na levoj strani promene. On je povezivao čir sa oštrim kvržicama donjih zuba, koje su gotovo uvek imale kontakt sa nastalim otokom u toku mastikacije. Pacijent nije primetio iscedak ili krvarenja iz otoka. Inače, pacijent je od ranije poznat klinici, jer boluje od dijabetesa melitusa tipa 2, koji je kontrolisao antidijabetičnom terapijom poslednjih pet godina.

Kliničkim pregledom evidentiran je usamljeni otok kupastog oblika leve strane maksile u distalnioj regiji, dimenzija približno 2 cm x 2 cm. Lezija se protezala anteriorno od zadnjeg kraja tubera maksile, zahvatajući meko nepce , medijalno nekoliko milimetara udaljeno od srednje linije , bočno zahvatajući bukalni vestibulum i posteriorno 2 cm udaljeno od tubera. Bočna strana otoka pokazala je prisustvo solitarnog tumefakta bez drenaže ili krvarenja. Prilikom palpacije, otok je bio čvrste konzistencije i neosetljiv (Slika 1). Razmatrana je diferencijalna dijagnoza rezidualne ciste u levoj zadnjoj maksilarnoj regiji, pošto je pacijent ukazao na istoriju ekstrakcije zuba iz tog predela.

Introduction

Pleomorphic adenoma is a benign neoplasm which is typically a non tender, slow growing mass and firm in consistency in majority of the cases. It has a slight female predilection. Although majority of the cases are associated with major salivary glands, palate is the common site for minor salivary gland pleomorphic adenomas[1].

Case Report

A patient presented to the Department of Oral Medicine and Radiology with a main complaint of swelling in the upper left back tooth region lasting 3 months. History revealed that the patient had visited a dentist for extraction of the upper left back tooth 5 months previously. Two months after the extraction, the patient noticed a very small swelling in that region which gradually during 3 months grew to the present size. This slow-growing swelling was associated with a dull, aching kind of pain which was intermittent and was non-radiating, with no aggravating and relieving factors associated with the pain. Two weeks prior to presenting to the Department of Oral Medicine and Radiology, the patient noticed a small ulcer in the left side of the swelling. He associated the ulcer with the sharp cusp of the lower teeth which would always come in contact with the swelling while chewing. There was no history of discharge or bleeding from the swelling. The patient was a known case of Type 2 Diabetes Mellitus with 5 years of medication use.

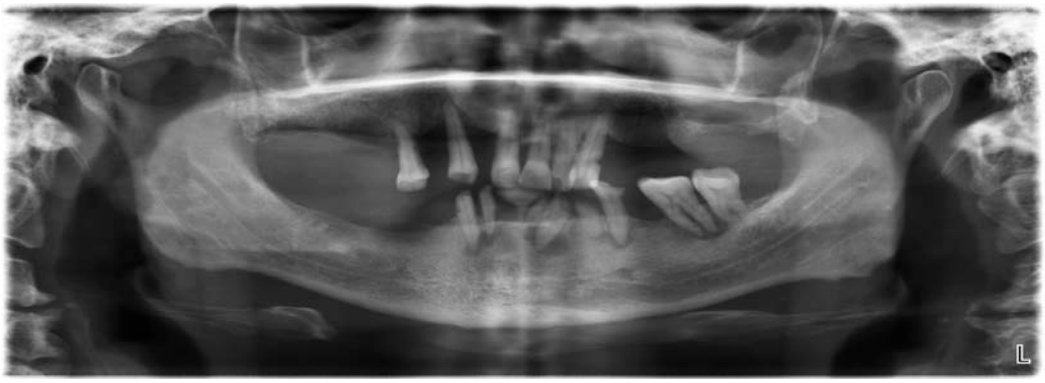
On clinical examination, a solitary dome-shaped swelling was evident in the left posterior maxillary region beyond the left maxillary tuberosity measuring ap-proximately 2 cm x 2 cm. The lesion extended anteriorly from the posterior end of the left maxillary tuberosity involving the soft palate, medially a few millimetres away from the midline, laterally involving the buccal vestibule and posteriorly 2 cm away from the tuberosity. The lateral side of the swelling showed the presence of solitary ulcer with no discharge or bleeding.The swelling was firm in consistency and non-tender on palpation (Figure 1). A provisional diagnosis of residual cyst in the left posterior maxilla was considered as the patient gave the history of having the tooth extraction done in the same region.

Slika 1. Usamljeni otok kupastog oblika leve distalne maksilarne regije

Figure 1. A solitary dome-shaped swelling was evident in the left posterior maxillary region

Na panoramskom rendgen snimku uočeno je rasvetljenje u levoj zadnjoj maksilarnoj regiji sa destrukcijom i resorpcijom alveolarnog nastavka. Radiografija je takođe otkrila generalizovanu apikalnu migraciju interdentalne kosti do apikalne trećine korena, što ukazuje na teški parodontitis. Uočeno je prisustvo loše definisane radiolucencije oko vrhova korenova zuba 31, 32, 33, 34, 36, 42 i 46, što ukazuje na hronične periapikalne apscese (Slika 2). Razmatrana je radiografska diferencijalna dijagnoza benignih odontogenih/ neodontogenih cista ili tumora, benignih malih tumora pljuvačnih žlezdi. Tumori mekog tkiva poput fibroma takođe su bili smatrani mogućom diferencijalnom dijagnozom.

Panoramic radiograph revealed a shadow of a soft tissue swelling in the left maxillary posterior region with destruction and resorption of the alveolar process. The radiograph also revealed generalised apical migration of the interdental bone up to the apical third of the roots suggestive of severe periodontitis. There was the presence of ill-defined radiolucency surrounding the apex of 31, 32, 33, 34, 36, 42 and 46 suggestive of chronic periapical abscesses (Figure 2). Radiographic differential diagnosis of benign odontogenic/non-odontogenic cysts or tumours, benign minor salivary gland tumours were considered. Soft tissue tumours like fibroma were also considered as a possible differential diagnosis.

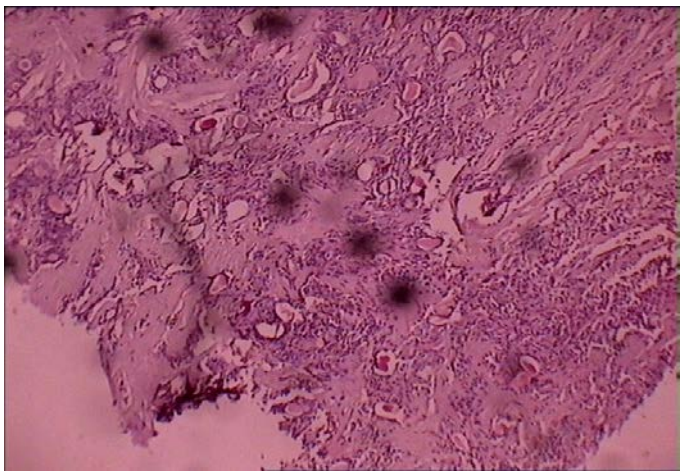


Slika 2. Panoramski rendgenski snimak koji otkriva senku otoka mekog tkiva u levoj zadnjoj maksilarnoj regiji sa destrukcijom i resorpcijom alveolarnog nastavka

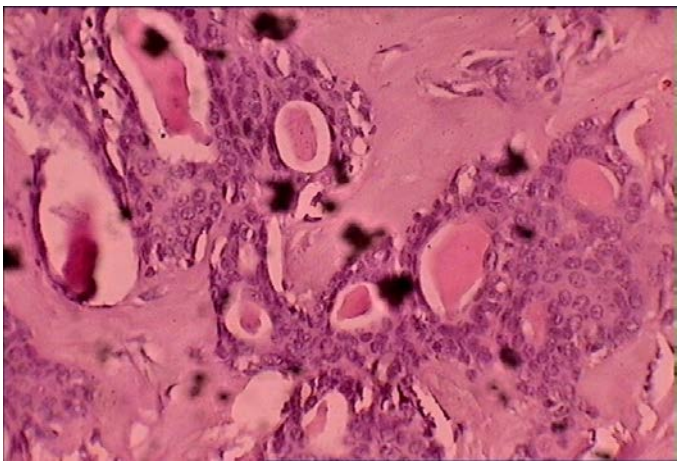
Figure 2. Panoramicradiographrevealing a shadow of a soft tissue swelling in the left maxillary posterior region with destruction and resorption of the alveolar process

Urađena je eksciziona biopsija i lezija je uklonjena u celini do periosta, a zatim je defekt potpuno prekriven sluzokožom. Uzorak je poslat na histopatološki pregled. Histopatološki pregled obojenih preseka pokazao je uglavnom ćelijske i hijalinizovane oblasti. Ćelije su bile epitelnog porekla, uglavnom okruglog oblika, sa nekoliko ćelija u obliku vretena, što je ukazivalo na to da su mioepitelnog porekla. Okrugle ćelije sadržale su vezikularna jedra. Uočene su strukture slične kanalima, koje sadrže eozinofilni koagulum ili mukoidni materijal. Neke oblasti koje okružuju vretenaste ćelije sastoje se od rastresite ili miksoidne strome . Vlakna kolagena, koja okružuju samu leziju primećena su u nekoliko oblasti , što ukazuje na fibroznu kapsulu. Uočena su i žarišta mukoznih acinusa, krvnih sudova, neurovaskularnog snopa i žarišta površinskog epitela. Ove karakteristike upućivale su na pleomorfni adenom (Slika 3 i Slika 4).

Excisional biopsy was carried out and the lesion was removed in total up to the periosteum and the overlying mucosa. The sample was sent for histopathological examination. Histopathological examination of the H and E stained sections showed mainly cellular and hyalinized areas. The cells were epithelial in origin, mainly round in shape with few spindle shaped cells suggesting their myoepithelial origin. The round cells contained vesiculated nuclei. Duct like structures containing eosinophilic coagulum or mucoid material were seen. Some areas surrounding the spindle cells consisted of loose or myxoid stroma. Collagen fibres surrounding the lesion proper were seen in a few areas, suggesting it to be the fibrous capsule. Foci of mucous acini, blood vessels, neurovascular bundle and foci of surface epithelium were also seen. These features were suggestive of Pleomorphic adenoma (Figure 3 and 4).



Slika 3. Histopatološki prikaz biosiranog materijala
Figure 3. Photomicrograph of H and E stained sections



Slika 4. Histopatološki prikaz biopsiranog materijala
Figure 4. Histopathological presentation of biopsied material

Diskusija

Pleomorfni adenom je tipično asimptomatski, spororastući tumor, uglavnom solidne konzistencije. Žene su unekoliko podložnije razvoju ove promene. U našem slučaju pacijent je muškog pola. U studiji Pitagaro i sar.[2] navodi se da je tumor detektovan kod 16 žena starosti između 16-30 god. i kod samo 4 osobe muškog pola, od ukupno 20 uključenih u studiju. Iako se u većini slučajeva javlja u velikim pljuvačnim žlezdama, smatra se da je nepce takođe predilekciono mesto za manje pleomorfne adenome pljuvačnih žlezda. Ovaj tumor je obično glatke površine, kupastog oblika, dobro ograničen i inkapsuliran[1].Ovakva lokalizacija je prisutna i u našem prikazu slučaja. Postojala je oteklina u obliku kupole koja je takođe pokazivala znake ulceracije. Patigaroo i sar.[2] su primetili da je samo 15% otoka pokazalo ulceraciju, 85% je pokazalo normalnu prekrivenu sluzokožu, 90% je imalo glatki otok, a 10% je pokazalo lobulirane otoke. Pored toga, nijedan slučaj nije pokazao recidiv u periodu praćenja[2].

Tumor je mešavina žlezdanog epitela i mioepitelnih ćelija unutar mezenhimalne pozadine. Hirurška ekscizija je tretman izbora. Ako tumori zahvataju tvrdo nepce, kao u našem sadašnjem slučaju, lezija se potpuno uklanja do periosta, sa uključivanjem sluznice koja pokriva leziju.

Pleomorfni adenom ima veoma dobru prognozu i beleži stopu izlečenja veću od 95%. Tumori malih pljuvačnih žlezda imaju veoma nisku stopu recidiva. Kada je hirurška resekcija urađena u potpunosti, skoro da ne postoji mogućnost recidiva. Recidiv se javlja kao posledica neadekvatne hirurške ekscizije[3]. Pleomorfni adenomi sa pretežno miksoidnim izgledom podložniji su recidivu[4,5].

Zaključak

Pleomorfni adenom je retko stanje. Rana dijagnoza i pravovremeni tretman su sinonimi za odličnu prognozu . Iako su stope niske, ne mogu se isključiti recidivi i maligna transformacija u dužem vremenskom periodu, posebno oni sa miksoidnim izgledom. Stoga je periodično dugoročno praćenje izuzetno obavezno za sve slučajeve.

Zahvalnica: Nema

Sukob interesa: Nema

Discussion

Pleomorphic adenoma is typically a non-tender slow-growing mass which is firm in consistency in majority of the cases. It is a benign neoplasm with slight female preponderance. In our case, the patient affected is a male, despite the female predilection stated in literature. In a study conducted by Patigaroo et al., 16 males were affected, predominantly in the age group of 16-30 years, and there were only 4 females out of the 20 people included in the study[2]. Although it occurs in the major salivary glands on most occasions, palate is considered to be a common site for minor salivary gland pleomorphic adenomas. This tumour is usually smooth surfaced, dome-shaped, well-circumscribed and encapusalated[1]. Further, in our case, the palate was the affected site. There was a dome-shaped swelling which also showed signs of ulceration. Patigaroo et al. observed that only 15% of the swelling showed ulceration, 85% showed normal overlying mucosa, 90% had a smooth swelling and 10% showed lobulated swellings. Additionally, no cases showed recurrence in the follow up period[2].

This tumour is a mixture of glandular epithelium and myoepithelial cells within the mesenchymal background. Surgical excision is the treatment of choice. If the tumours involve the hard palate as in our present case, the lesion is completely excised down to the periosteum with the inclusion of the overlying mucosa.

Pleomorphic adenoma has a very good prognosis and records a cure rate of more than 95%. Tumours of the minor salivary glands have very low recurrence rates. If the surgical excision is adequate, chances of recurrence are minimal. Recurrence can be associated with inadequate surgical excision[3]. Pleomorphic adenomas with predominantly myxoid appearance are more susceptible to recurrence[4,5].

Conclusion

Pleomorphic adenoma is a rare condition. Early diagnosis and prompt treatment are synonymous with an excellent prognosis. Although the rates are low, recurrence and malignant transformation over a long period of time cannot be ruled out, especially the ones with myxoid appearance. Hence periodic long term follow up is extremely mandatory for all cases.

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Conflict of interest: Nil

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