Mioepiteliomi su benigné neoplazme žlijezda slinovnica i uglavnom se sastoje od stanica mioepitelnog fenotipa ili većinom od tih stanica (1, 2). Pojavljuju se s približno 1,5 posto tumora žlijezda slinovnica (3) i obično zahvaćaju parotidu ili malo žlijezda slinovnice na nepcu (1–4). Nema preferencije u dobi ili spolu, ali češće obolijevaju osovine (1–4). Obično se pojavljuju asimptomatski, dobro ograničena, neoplazma, ali nisu učahurena, i ne i učahurena, i nisu i normalne su boje, na dodir su meke do tvrde (2–4). Obično im se pojavljuju simptomatski, dobro ograničene tumorske tvorbe koje rastu sporo u labavoj fibrovaskularnoj stromi. Imunohistokemijsko je vidljivo da se plazmacitoidni mioepiteliom pojavljuje, međutim ovo nije bio uočeno vracanje novotvorine. Zaključak: Budući da se plazmacitoidni mioepiteliom pojavljuje, njegova imunohistokemijska svojstva, liječenje i prognoza trebali bi se još detaljnije istražiti.

Uvod

Myoepitheliomas are benign salivary gland neoplasms consisting entirely or predominantly of cells with myoepithelial phenotype (1, 2). They represent approximately 1.5% of all salivary glands tumors (3) and usually involve the parotid gland or the minor salivary glands of the palate (1–4). They show no age or gender predilection, but they are more common in middle-aged persons (1–4). They present as asymptomatic, slowly growing and well-circumscribed tumors of normal color that may be soft to hard on palpation (2–4), and are not associated with neurological symptoms (3, 4).

Microscopically, one or all types of neoplastic myoepithelial cells, i.e. spindle, plasmacytoid (hyaline), epithelioid, clear, polygonal, basoloidal or oncocytic, may be seen, arranged in solid, myxoid, reticular, microcystic or cribriform growth patterns (1, 3, 5). The preponderant cell subtype defines the tumor’s subtype, although neither cell type nor growth pattern correlate with the clinical presentation or biologic behavior of the lesion (1). The spindle cells subtype is more common in the parotid gland and the plasmacytoid in the minor salivary glands of the palate (2). Whether myoepithelioma is a distinct entity or a variant of pleomorphic adenoma with a preponderance of cells with myoepithelial phenotype is disputable.

Since the review of the English literature by Zormpa et al (6) in 2011, where 19 cases of plasmacytoid myoepithelioma of the hard palate were included, three more cases have been published (7,8,9). An additional case of plasmacytoid myoepithelioma of the hard palate is described.

Plazmacitoidni mioepiteliom tvrdog nepca: prikaz slučaja

Plasmacytoid Myoepithelioma of the Hard Palate: Case Report

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Svuha

Svrha: Mioepiteliomi su rijetke neoplazme žlijezda slinovnica, a sastoj se od stanica mioepitelnog fenotipa. Obično zahvaćaju parotitidu i male žlijezde slinovnice na nepcu. Svrha rada: Opis: Opisan je slučaj plazmacitoidnog mioepitelioma tvrdog nepca. Opis slučaja i rezultati: Opisani tumor buknuo je na tvrdom nepcu 55-годишnje žene. Mikroskopskim pregledom ustanovljeno je dobro ograničeno neoplazma, ali ne i učahurena, a sadržavala je uglavnom plazmacitoidne stanice u labavo fibrovaskularnoj stromi. Imunohistokemijski te su stanice reagirale s proteinom s-100, CK AE1/AE3 i GFAP-om, kalponinom i CD138/sindekanom 1. Terapija se sastojala od uklanjanja cijelog tumora u lokalnoj anesteziji. Ni 14 mjeseci nakon zahvata nije bilo uočeno vracanje novotvorine. Zaključak: Budući da se plazmacitoidni mioepiteliom pojavljuje, njegova imunohistokemijska svojstva, liječenje i prognoza trebali bi se još detaljnije istražiti.

Uvod

Opis slučaja i rezultati:

Opisan je slučaj plazmacitoidnog mioepitelioma tvrdog nepca 55-godišnje žene. Mikroskopskim pregledom ustanovljena je uglaovnom plazmacitoidna stanica, primjerice vretenasta, plazmacitoidna (hyaline), epithelioidna, čista, poligonalna, bazaloidna ili oncocitna i sve su poredane jednolično prema miksoidnom, idne (hijaline) epiteloidne, čiste, poligonalne, bazaloidne ili oncocitne.

Mikroskopski je vidljiv jedan tip ili svi tipovi neoplastičnih mioepitelnih stanica, primjerice vretenaste, plazmacitoidne (hijaline) epiteloidne, čiste, poligonalne, bazaloidne ili oncocitne, ali nisu asocijirane s neurološkim simptomima (3, 4). Mikroskopski je vidljiv jedan tip ili svi tipovi neoplastičnih mioepitelnih stanica, primjerice vretenaste, plazmacitoidne (hijaline) epiteloidne, čiste, poligonalne, bazaloidne ili oncocitne, ali nisu asocijirane s neurološkim simptomima (3, 4). Mikroskopski je vidljiv jedan tip ili svi tipovi neoplastičnih mioepitelnih stanica, primjerice vretenaste, plazmacitoidne (hijaline) epiteloidne, čiste, poligonalne, bazaloidne ili oncocitne, ali nisu asocijirane s neurološkim simptomima (3, 4). Mikroskopski je vidljiv jedan tip ili svi tipovi neoplastičnih mioepitelnih stanica, primjerice vretenaste, plazmacitoidne (hijaline) epiteloidne, čiste, poligonalne, bazaloidne ili oncocitne, ali nisu asocijirane s neurološkim simptomima (3, 4).

Zaključak:

Budući da se plazmacitoidni mioepiteliom trijetko pojavljuje na žlijezdama slinovnicama, njegova imunohistokemijska svojstva, liječenje i prognoza trebali bi se još detaljnije istražiti.
Case report

A 55 year-old woman was referred for diagnosis and management of a painless swelling on the hard palate that had gradually enlarged during the last few months. Family and past medical histories were non-contributory.

Oral examination revealed a round, well-circumscribed mass covered by normal mucosa on the right posterior hard palate, between the premolar teeth and the midline (Figure 1). It measured approximately 2.5x2x1.5 cm and was compressible and non-tender on palpation. The first molar tooth did not react to pulp testing, but adjacent teeth were vital.

Prikaz slučaja

Žena od 55 godina upućena je na dijagnosticiranje i terapiju bezbolne otekline na tvrdom nepcu koja se postupno povećavala tijekom nekoliko mjeseci. U osobnoj i obiteljskoj anamnezi nije bilo takve bolesti.

Oralnim pregledom otkrivena je okrugla, dobro ograničena masa prekrivena normalnom služnicom na desnom stražnjem dijelu nepca, između pretkutnjaka i središnje linije (slika 1.). Bila je velika oko 2,5 x 2 x 1,5 centimetara, mogla se stisnuti i bila je mekana na palpaciju. Prvi kutnjak nije reagirao na testiranje pulpe, ali ostali susjedni zubi bili su vitalni. Nije

Slika 1. Oralni pregled – okrugla, dobro ograničena masa prekrivena normalnom služnicom na stražnjem desnom dijelu tvrdog nepca, između pretkutnjaka i središnje linije

Figure 1 Oral examination. A round, well-circumscribed mass covered by normal mucosa on the right posterior hard palate, between the premolar teeth and the midline.

Slika 2. Zubni sken – prosvjetljenje mase koja ne zahvaća maksilarnu kortikalnu kost; cistična lezija vidljiva je apikalno od prvoga kutnjaka (zvjezdica)

Figure 2 Dental scan. The hypodense mass does not involve the maxillary cortical bone. A cystic lesion is seen apically to the first molar tooth (asterisk).

Slika 3. Kontrola – poslijeoperacijsko cijeljenje 14 mjeseci nakon terapije

Figure 3 Follow up. Postsurgical healing 14 months after treatment.

Slika 4. Mikroskopska obilježja – (a) solitarni čvorovi neoplastičnih stanica uklonjenih labavo u vaskularnu stromu vezivnoga tkiva (hematoksilin-eozinsko bojenje, povećanje 200 x); (b) plazmacitoidne i (c) vrtenaste mioepitelijalne stanice (hematoksilin-eozinsko bojenje, povećanje 400 x).

Figure 4 Microscopic features. (a) Solid nests of neoplastic epithelial cells embedded in a loose vascular connective tissue stroma (hematoxylin and eosin stain, original magnification x200). (b) Plasmacytoid and (c) spindle-shaped myoepithelial cells (hematoxylin and eosin stain, original magnification x400).

Slika 5. Imunohistokemijska obilježja plazmacitoidnih stanica – (a) intenzivno pozitivna reakcija u citoplazmi na protein S-100 i (b) na GFAP (c); snažna pozitivna reakcija na membrani za CD138/Syndecan-1 (originalno povećanje 400 x)

Figure 5 Immunohistochemical features of plasmacytoid cells. (a) An intense cytoplasmic positivity for S-100 protein and (b) GFAP. (c) Strong, membranous positivity for CD138/Syndecan-1 (original magnification x400).
Mikroskopskim pregledom rezova od 5 μm tkiva fiksiranih u formalinu (urojenih u parafin), uočena je dobro ograničena tvorba bez čažure koja se sastojala od slojeva i nakupina neoplastičnih epitelnih stanica uklupljenih u labavu fibrovaskularnu stromu (slika 4. a). Većina stanica imala je plazmacitoidna (hijalina) miliohidna obilježja, primjerice obilnu eozinofilnu neoplazmu i ovalnu, blagu zgusnutu, eksenčričnu jezgru (slika 4. b). Uočeno je i mala grupa vretenastih stanica s gustim jezgroma (slika 4. c) te minimalni stanični i nuklearni pleomorfizam te bez netipičnih mitoza. Zabilježene su i rijetke strukture odvodnih kanala koje su činile samo dva posto ukupnog parenhima, a acinusna diferencijacija je nedostajala. Na mjestu inicijalne biopsije pada se reaktivni izraz u formi plazmacitoidnih mioepitelioma.

Immunohistochemistry was performed with a standard avidin-biotin peroxidase technique after pretreatment with high temperature citrate buffer, with antibodies against α-SMA and p63, and strong, membranous positivity for S-100 protein (Figure 5a), and plasmacytoid and spindle cells showed intense cytoplasmic positivity for α-SMA and p63, as well as Ki-67 proliferation index Ki-67 (1, 50 μm thick formalin-fixed and paraffin-embedded tissue sections showed a well-circumscribed but non-encapsulated tumor, consisting of solid sheets and nests of neoplastic epithelial cells embedded in a loose fibrovascular stroma (Figure 4a). Most cells showed plasmacytoid (hyaline) myoepithelial features, i.e. abundant eosinophilic neoplasm and an oval, slightly dense, eccentric nucleus (Figure 4b). Small groups of spindle-shaped cells with dense nuclei were also observed (Figure 4c). There was a minimal cellular and nuclear pleomorphism and there were no atypical mitoses. Rare ductal structures were seen, but constituted <2% of the total tumor parenchyma, while acinar differentiation was absent. Hemorrhage, inflammation and pseudopseudotumorous hyperplasia of the covering parakeratinized mucosa were seen in the site of the incisional biopsy.

Discussion

The case presented herein showed clinical, microscopic and immunohistochemical features consistent with plasmacytoid myoepithelioma. Panoramic radiograph and dental scan found no sign of erosion of the adjacent bone that, however, may be seen in benign salivary gland tumors, including plasmacytoid myoepithelioma (7). Microscopically, differentiation from a common pleomorphic adenoma was based on limited ductal differentiation that in our case was <2%, lack of chondroid or osteoid stroma, and predominance of myoepithelial cells (1, 4).
The immunophenotype of neoplastic cells in myoepitheliomas may vary among different tumors (1, 2, 5) and among different cell types of the same tumor (1, 2, 5). In our case, both plasmacytoid and spindle cells were S-100 positive, and plasmacytoid cells, unlike spindle cells, showed diffuse cytoplasmic positivity for CK AE1/AE3, GFAP, and calponin. Expression of those antibodies in plasmacytoid myoepithelioma is a constant finding (6, 14), while SMA and GFAP positivity may vary (6, 7, 14). In our case, α-SMA expression was very limited, but GFAP expression was diffuse. GFAP expression could possibly facilitate differentiation from myoepithelial carcinoma, as neoplastic myoepithelial cells of pleomorphic adenoma are GFAP positive, but of polymorphous low-grade adenocarcinoma GFAP negative (11). Limited p63 expression was also seen in our case and p63 was found in some cutaneous myoepitheliomas (15) and salivary myoepithelial carcinomas (16,17), therefore, its application in differentiation of plasmacytoma from its malignant variant should be further evaluated. CD138/Syndecan-1 is expressed by normal and neoplastic plasma cells, as well as neoplastic cells with plasmacytoid features (18). Strong, membranous positivity for CD138/Syndecan-1, as seen in the present case, was reported in a single case of plasmacytoid myoepithelioma (18). Lack of cytological atypia, cellular pleomorphism, necrosis, hemorrhage, infiltration into adjacent tissue, as well as low mitotic rate were highlighted by Ki-67 labeling index of <1% and they precluded a diagnosis of malignant myoepithelioma (1, 2, 10).

Complete surgical excision on tumor-free margins is usually curative (2), although myoepithelioma is reported to have more aggressive behavior to pleomorphic adenoma (10). Recurrence is associated with positive margins and this may occur in myoepitheliomas of minor salivary glands, especially of the palate, where tumor encapsulation is uncommon (1). Recurrence rate of plasmacytoid myoepitheliomas of the palate is estimated to 15-18% (6,14). Malignant transformation may ensue, particularly in the spindle cell variant (12).

Conclusions

Since plasmacytoid myoepithelioma is an uncommon tumor of minor salivary glands, its immunohistochemical features, management and prognosis should be further investigated through the publication of more documented cases.

Zaključak

Budući da je plazmacitoidni mioepiteliom neubojčen tumor malih žlijezda slinovnica, njegova imunohistokemijska obilježja, terapija i prognoza trebali bi se dalje istraživati, dobiveni rezultati dokumentirati i objavljivati.

Sukob interesa

Autori izjavljuju da nisu u bili u sukobu interesa.
Abstract

Background: Myoepitheliomas are uncommon salivary gland neoplasms consisting entirely or predominantly of cells with myoepithelial phenotype. They commonly involve the parotid gland and the minor salivary glands of the palate. Aim: A case of plasmacytoid myoepithelioma of the hard palate is described. Case description and results: A 55-year-old woman presented to her oral surgeon with a tumor on the hard palate. Microscopic examination showed a well-circumscribed but non-encapsulated tumor, consisting mostly of plasmacytoid cells in a loose fibrovascular stroma. Neoplastic myoepithelial cells showed immunoreactivity for S-100 protein, CK AE1/AE3 (Figure 5b), GFAP, calponin, and CD138/Syndecan-1. Total excision of the tumor under local anesthesia was performed and no recurrence was noted 14 months after treatment. Conclusions: Since plasmacytoid myoepithelioma is uncommon, minor salivary glands, its immunohistochemical features, management and prognosis should be further investigated.

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References

1. Gnepp DR. Diagnostic Surgical Pathology of the Head and Neck. 2nd ed. Philadelphia: Saunders; 2001.
2. Hunt KT, Stevens MR, Abdelsayed RA, Nguyen CT. Benign myoepithelioma of floor of mouth with mandibular involvement: a case report and literature review. J Oral Maxillofac Surg. 2011 Dec;69(12):2301-5.
3. Ellis AL, Auclair PL. Benign epithelial neoplasms. 3rd ed. Washington, DC: Armed Forces Institute of Pathology; 1996.
4. Barnes L, Appel BN, Perez H, El-Attar AM. Myoepithelioma of the head and neck: case report and review. J Surg Oncol. 1985 Jan;28(1):21-8.
5. Savera AT, Zarbo RJ. Defining the role of myoepithelioma in salivary gland neoplasia. Adv Anat Pathol. 2004 Mar;11(2):69-85.
6. Zormpa MT, Sarigelou AS, Eleftheriou AN, Assimaki AS, Kolokotronis AE. Plasmacytoid myoepithelioma of the palate: case report. Head Neck Pathol. 2011 Jun;5(2):154-8. 3098329.
7. Santos EP, Cavalcante DR, Melo AU, Pereira JC, Gomes MZ, Albuquerque RJ Jr. Plasmacytoid myoepithelioma of minor salivary glands: report of case with emphasis in the immunohistochemical findings. Head Face Med. 2011 Dec 12;7:26.
8. Sethi D, Ahluwalia C, Kathri A, Khetarpal S. Palatal plasmacytoid myoepithelioma. Adv Biomed Res. 2012;1:78.
9. Kulkarni PR, Javalgi AP, Pottipati B, Shajahan F4. Plasmacytoid Myoepithelioma of the Hard Palate in a Child - A Rare Case Report. J Clin Diagn Res. 2015 Oct;9(10):ED01-2.
10. Hornick JL, Fletcher CD. Cutaneous myoepithelioma: a clinicopathologic and immunohistochemical study of 14 cases. Hum Pathol. 2004 Jan;35(1):14-24.
11. Curran AE, Allen CM, Beck FM, Damm DD, Murrah VA. Distinctive pattern of glial fibrillary acidic protein immunoreactivity useful in distinguishing fragmented pleomorphic adenoma, canalicular adenoma and polymorphous low grade adenocarcinoma of minor salivary glands. Head Neck Pathol. Head Neck Pathol. 2007 Sep;1(1):27-32.
12. Alos L, Cardesa A, Bomba JA, Mallofre C, Cuchi A, Traserra J. Myoepithelial tumors of salivary glands: a clinicopathologic, immunohistochemical, ultrastructural, and flow-cytometric study. Semin Diagn Pathol. 1996 May;13(2):138-47.
13. Gore CR, Panicker NK, Chandanwale SS, Singh BK. Myoepithelioma of minor salivary glands- A diagnostic challenge: Report of three cases with varied histomorphology. J Oral Maxillofac Pathol. 2013 May;17(2):257-60.
14. Kulkarni PR, Javalgi AP, Pottipati B, Shajahan F. Plasmacytoid Myoepithelioma of the Hard Palate in a Child - A Rare Case Report. J Clin Diagn Res. 2015 Oct;9(10):ED01-2.
