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Case Report

Multifocal nodular oncocytic hyperplasia clear cell variant: a rare case report of salivary gland lesion

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Abstract

Nodular oncocytic hyperplasia is rare non-neoplastic lesion of the salivary glands. Histopathologically, it is characterized by multiple unencapsulated nodular proliferations. It can be confused with tumors in the differential diagnosis both cytopathologically and histopathologically. Differential diagnosis should be made with metastatic tumors and clear cell salivary gland tumors, especially if clear cells predominate. In this rare case, we presented the cytological, histopathological and immunohistochemical findings of clear cell dominant nodular oncocytic hyperplasia in the unilateral parotid deep lobe in a 72-years-old male patient.

Keywords: Nodular oncocytic hyperplasia, oncocytosis, salivary gland.

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INTRODUCTION

Nodular oncocytic hyperplasia is among the non-neoplastic lesions of the salivary glands in the latest World Health Organization (WHO) classification (1). It has definitions such as multifocal nodular oncocytic hyperplasia and clear cell oncocytosis (1). Histopathologically, they are multiple nodular lesions consisting of oncocytes and/or clear cells in the natural salivary gland (1). Since they form masses and their cytological diagnosis is difficult, they are diagnosed in excision materials. In the light of our findings, we wanted to present our elderly male patient with unilateral right parotid deep lobe involvement with clear cell predominance different than expected.

CASE REPORT

A 72-years-old male patient was admitted to the hospital with complaints of swelling in front of the right ear, tinnitus and hearing loss for one year. A palpable and semimobile mass of approximately 2x1.5 cm was observed on the right angulus mandible. Ultrasound examination revealed a 12x9 mm hypoechoic solid nodule with lobulated contour and central hemorrhage in the right parotid deep lobe. Lesions compatible with lymph node, the largest of which is 9x7 mm, were observed in other areas. On Magnetic Resonance Imaging (MRI), lesions consistent with multiple lymph nodes were observed in both parotids, the largest on the right, measuring 12x9 mm. Thin-needle aspiration biopsy showed cells with narrow-medium-wide cytoplasm, generally uniform round nuclei, fine granular chromatin, small nucleolus, which form monolayer and papillary structures on the hemorrhagic background, occasionally cohesive and some distributed in three-dimensional groups. The cytology report was reported as a salivary gland neoplasm of uncertain malignant potential (SUMP). Right paratidectomy was planned for the patient. After the operation, 8x6x2 cm gray colored paratidectomy material was sent. When sliced, a nodular lesions, the largest of which was 11x10 mm in size, homogeneous on the incision surface, gray in color, and well-circumscribed was observed. Numerous well-circumscribed nodular lesions in different sizes were observed in other areas of the parenchyma. In the examination of the preparations, proliferations was observed in the parenchyma, which developed in a nodular manner in many different diameters and sizes, well-circumscribed, occasionally cystic areas and vascular-rich reticulated stroma. The lesions consisted of cells with large clear cytoplasm in most areas, eosinophilic cytoplasm in some areas, and round-oval nuclei with discernible nucleoli in places (Figure 1). No lesion was observed in the parenchyma in other areas. In the immunohistochemical examination, these cells were Pan-cytokeratin (PANCK), CK7, p63 (+); PSAP, S100, SMA, CD10, Vimentin, RCC, CK20, PAX8 (-) were detected (Figure 2). CD10 and vimentin were positive in the stroma. The proliferation index with Ki67 was less than 1%. Staining with PAS, PAS-D, mucin was not observed. Mitosis, atypia, necrosis were not observed. No lesion was observed in the surgical margins. The case was followed for one year and no lesion was detected in the salivary glands.

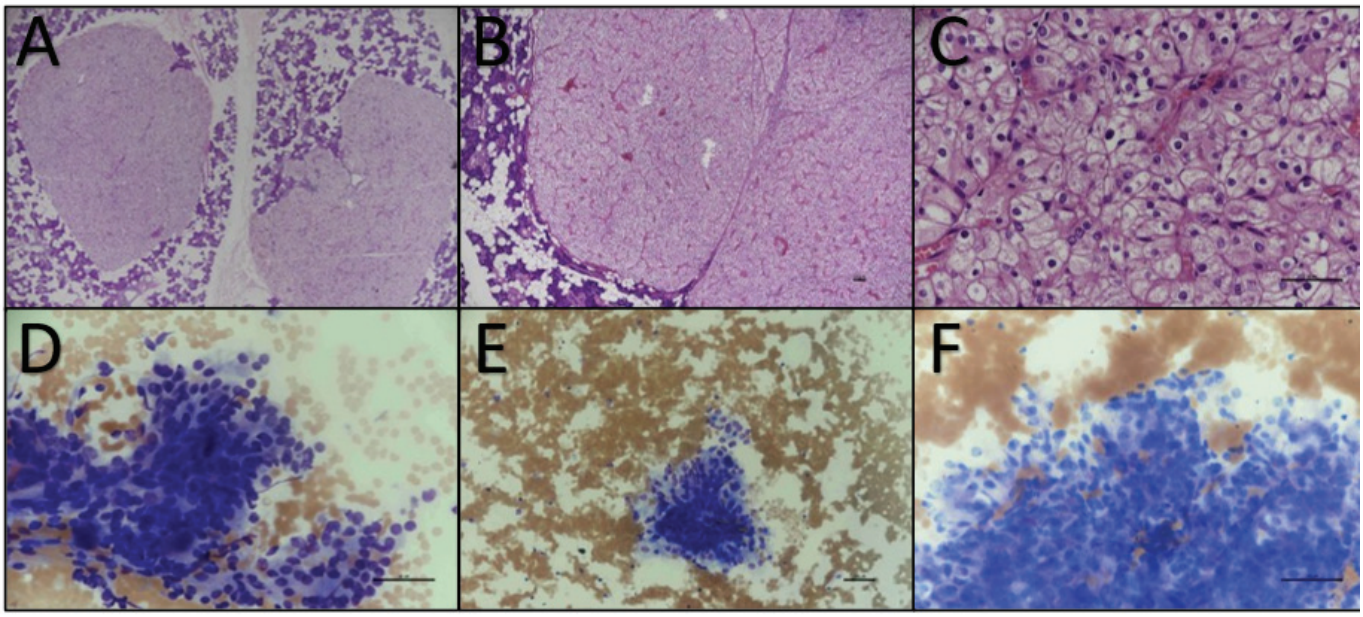

Figure 1. A, B: Well circumscribed nodular proliferation in normal salivary gland parenchyma (HE X2, HE X4). **C**: High power view of cell with clear cytoplasm centrally round nuclei (HE X40). **D, E, F**: Cytological features; cells with narrow-medium width cytoplasm, generally uniform round, fine granular chromatin, small nucleolus can be distinguished, forming monolayers and papillary structures in between and distributed in cohesive three-dimensional groups (HE X10, X4, X40).

DISCUSSION

Nodular oncocytic hyperplasia was first described by Schwartz (2). Nodular oncocytic hyperplasia is a nonneoplastic lesion that is not expected to become malignant (1). It is observed around the age of 57 on average and generally in women (1). It is seen in the parotid gland and is bilateral in 40% of cases (1). Recurrence can be expected in cases that cannot be completely removed surgically (1). The lesion may sometimes accompany other tumors. Cases occurring together with oncocytoma and after pleomorphic adenoma have been reported (3,4). Cytologically, it is described as clusters of oncocytic cells with reduced nucleus/cytoplasm ratio, central nuclei, prominent nucleoli (5). With these cytological features, lesions such as Warthin tumor, oncocytoma, papillary oncocytic cystadenoma/cystadenocarcinoma, salivary duct carcinoma, acinic cell carcinoma are included in the differential diagnosis (6). In our case, due to the predominance of oncocytic cells in cytology, surgical resection was performed to exclude the tumoral lesion. When we examined it after resection, we made a differential diagnosis with metastatic tumors because histopathologically it developed as multifocal nodules predominantly from clear cells. Like our case, the majority of oncocytes could be clear cells and immunohistochemical staining may be needed for differential diagnosis. Zhou et al. and Giordano et al. find their immunohistochemical properties useful in malignant cases (7,8). Metastatic renal cell carcinoma is stain with CD10, vimentin, and renal cell carcinoma antigen (9). In our case, the staining of metastatic renal cell carcinoma and other clear cell carcinomas was negative. In benign tumoral lesions, on the other hand, morphological analysis is based on, but immunohistochemical studies may be needed in necessary cases (10). Clear cell neoplasms that come to mind in clear cell lesions: oncocytoma, clear cell myoepithelioma (9). The presence of capsule is especially determinant in its differentiation from oncocytoma (9). In our case, no capsule was observed around the lesion. Nodular oncocytic hyperplasia can also be distinguished from epithelial-myoepithelial carcinoma and clear cell myoepithelioma by using immunohistochemical myoepithelial markers: cytokeratin 5/6 (CK5/6), cytokeratin 14 (CK14), p63 protein, vimentin (VIM), α-smooth muscle actin (Alpha-SMA), calponin and S-100 (9). Typical immunohistochemical staining for salivary gland tumors was not observed in our case. With these results, we evaluated our diagnosis as multifocal nodular oncocytic hyperplasia. Since our case was unilateral and the lesion could be seen bilaterally, it may be necessary to follow the other gland in unilateral cases (11).

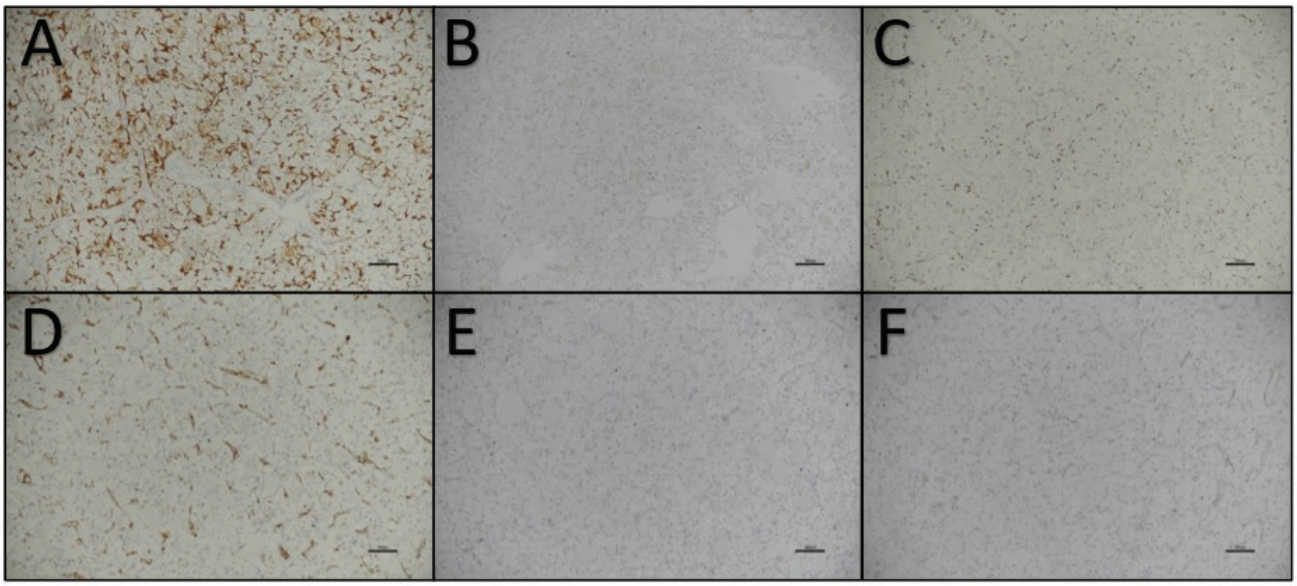

Figure 2. Immunohistochemical markers **A**: PANCK **B**: RCC **C**: p63 **D**: CD10 **E**: SMA **F**: S100 (X20).

CONCLUSION

We present the features of the clear cell type of multinodular oncocytic hyperplasia, which is a rare non-neoplastic lesion of the salivary gland. Malignantization is not expected in these lesions and surgical intervention is not required if the typical features are known. They are lesions that should be kept in mind in the differential diagnosis, especially as they are confused with renal cell carcinoma metastasis, especially if they consist of clear cells, as in our case.

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References

1. El-Naggar AK, Chan J, Takata T, Grandis J, Slootweg P. WHO Classification of tumors. In: Pathology and Genetics of Head and Neck Tumors. Lyon: IARC Press; 2017.
2. Schwartz IS, Feldman M. Diffuse multinodular oncocytoma ('oncocytosis') of the parotid gland. Cancer 1969;23(3):636–40.
3. Shellenberger TD, Williams MD, Clayman GL, Kumar AJ. Parotid gland oncocytosis: CT findings with histopathologic correlation. AJNR Am J Neuroradiol 2008;29(4):734–6.
4. Yue LE, Spaulding SL, Baik FM, Khorsandi A, Brandwein-Weber M, Urken ML. Multifocal Parotid Oncocytic Cystadenomatosis: A Rare Process that Can Mimic Malignancy. Head Neck Pathol. 2021;15(1):323-7.
5. Goyal R, Ahuja A, Gupta N, Singh G, Vaiphei K. Multifocal nodular oncocytic hyperplasia in parotid gland: a case report. Acta Cytol. 2007;51(4):621-3.
6. Dardick I, Birek C, Lingen MW, Rowe PE. Differentiation and the cytomorphology of salivary gland tumors with specific reference to oncocytic metaplasia. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88(6):691-701.
7. Zhou CX, Shi DY, Ma DQ, Zhang JG, Yu GY, Gao Y. Primary oncocytic carcinoma of the salivary glands: a clinicopathologic and immunohistochemical study of 12 cases. Oral Oncol. 2010;46(10):773-8.
8. Giordano G, Gabrielli M, Gnetti L, Ferri T. Oncocytic carcinoma of parotid gland: a case report with clinical, immunohistochemical and ultrastructural features. World J Surg Oncol. 2006;4:54.
9. Sato S, Kishino M, Ogawa Y, Nakatsuka S, Hoshida Y, Ogawa I, et al. Multifocal nodular oncocytic hyperplasia of bilateral parotid glands: A case report with a histological variant of clear cells. Pathol Res Pract. 2011;207(7):452-5.
10. Rooper LM, Onenerk M, Siddiqui MT, Faquin WC, Bishop JA, Ali SZ. Nodular oncocytic hyperplasia: Can cytomorphology allow for the preoperative diagnosis of a nonneoplastic salivary disease? Cancer Cytopathol. 2017;125(8):627-34.
11. Akbulut A, Kaptan ZK, Yasaran M, Karadas H, Koca G, Korkmaz M. Metachronous bilateral multifocal nodular oncocytic hyperplasia of the parotid gland. J Stomatol Oral Maxillofac Surg. 2019;120(6):598-600.