

Case Report / Olgu Sunumu

Oncocytic lipoadenoma of the parotid gland: a case report with fine needle aspiration cytology findings

Parotis bezinde onkositik lipoadenom: İnce iğne aspirasyon sitolojisi bulguları ile birlikte bir olgu sunumu

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Oncocytic lipoadenoma of the salivary gland is a rarely encountered tumor. A 56-year-old man presented with a two-year history of a slow-growing mass of the left parotid gland. Computed tomography scan with contrast showed a 7x6.5x6 cm well-circumscribed solid parotid mass of the left superficial and deep lobe. Fine-needle aspiration yielded oncocytic cells exclusively, suggesting Warthin tumor or an oncocytoma. Left total parotidectomy was performed. A diagnosis of oncocytic lipoadenoma was made. At six-month follow-up no evidence of recurrence has been noted. Oncocytic lipoadenoma should be considered in the differential diagnosis of oncocytic proliferations and oncocytic tumors in the parotid gland.

Key Words: Lipoadenoma; oncocyte; parotid gland.

Tükürük bezinin onkositik lipoadenomu nadir görülen bir tümördür. Elli altı yaşında erkek hasta sol parotis bezinde iki yıldır yavaş büyüyen kitle öyküsü ile başvurdu. Kontrastlı bilgisayarlı tomografi incelemesinde sol süperfisyal ve alt derin lobda 7x6.5x6 cm boyutlarında solid iyi sınırlı parotis kitlesi saptandı. İnce iğne aspirasyon biyopsisinde görülen onkositik hücreler öncelikle Warthin tümörü veya onkositoma ön tanısını akla getirdi. Hastaya sol total parotidektomi uygulandı. Onkositik lipoadenom tanısı konuldu. Altı aylık takipte yineleme izlenmedi. Parotisdeki onkositik proliferasyonların ve onkositik tümörlerin ayırıcı tanısında onkositik lipoadenom da düşünülmelidir.

Anahtar Sözcükler: Lipoadenom; onkosit; parotis bezi.

Salivary gland tumors account for only 3% of all tumors in the body.^[1] They have a great variety of histological types. Oncocytic lipoadenomas are extremely rare tumors not included in the 2005 World Health Organization (WHO) histologic classification of tumors of the salivary glands.^[2] There are only four reported cases in the literature.^[3-6] We report an additional case with a description of the cytological appearance and other pathological findings.

CASE REPORT

A 56-year-old man presented with a two-year history of a painless, slow-growing mass of the left parotid gland. Computed tomography scan with contrast showed a 7x6.5x6 cm well-circumscribed solid parotid mass of the left superficial and deep lobe. Fine-needle aspiration yielded oncocytic cells exclusively, suggesting Warthin tumor or an oncocytoma (Figure 1).

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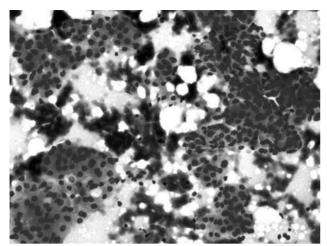


Figure 1. Cohesive groups of oncocytic cells with abundant cytoplasm (Giemsa, x 200).

The patient underwent a left total parotidectomy with preservation of the facial nerve. The surgical specimen consisted of a well circumscribed mass measuring 7 cm in maximum diameter. Microscopically, the the tumor was surrounded by a thin fibrous capsule, and was composed of an admixture of oncocytes, mature fat cells, ductal components, and sebacous glands (Figure 2, 3). Approximately 40% of the tumor was composed of adipose tissue, with the remaining being composed of oncocytes. The oncocytes showed abundant, granular cytoplasm. A second cell population composed of small densely eosinophilic cells, referred to as "dark cells", was also identified. There were scattered mature sebaceous glands admixed with the oncocytes. Some of the sebaceous glands were surrounded by small lymphoid aggregates (Figure 4). The adjacent parotid gland was

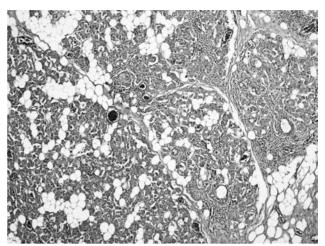


Figure 2. Admixture of oncocytes, mature fat cells, ductal component, and sebacous glands (H-E x 40).

unremarkable. Phosphotungstic acid hematoxylin stain demonstrated strong cytoplasmic granularity consistent with abundant mitochondria within the tumor cells. This confirmed their oncocytic nature. Immunohistochemically, the oncocytes were positive for keratin 7 and epithelial membrane antigen. Actin was negative, indicating a lack of myoepithelial cells. A diagnosis of oncocytic lipoadenoma was made.

At six-month follow-up, the patient did not report symptoms related to the parotid gland and no evidence of recurrence has been noted.

DISCUSSION

An oncocyte is an epithelial cell that probably originates from metaplasia of acinar or ductal epithelium.^[7] It is larger than an acinar cell and has an abundant granular eosinophilic cytoplasm.^[8]

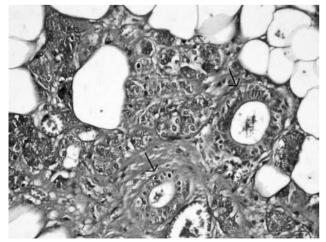


Figure 3. Ductal component at higher magnification (H-E x 200).

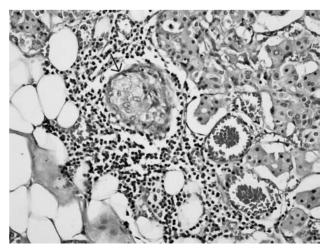


Figure 4. Sebaceous glands surrounded by small lymphocytes (H-E x 200).

Author	Age (year)/sex	Site	Size (cm)	Follow-up
Hirokawa et al. ^[3]	66/F	Submandibular gland	11.0	AND*, 30 months
Kato and Horie ^[4]	57/F	Parotid gland	4.5	NA**
Klieb and Perez-Ordoñez ^[5]	47/M	Parotid gland	3.8	AND*, 6 months
Aouad et al. ^[6]	38/M	Parotid gland	3.8	NA**
Current case	57/M	Parotid gland	7.0	AND*, 6 months

Table 1. Case reports of salivary gland oncocytic lipoadenoma

*: Alive no disease; **: Not available; F: Female; M: Male.

The cytoplasm is filled with tightly packed pleomorphic mitochondria. The cell's appearance may represent an intracellular age-related metabolic disturbance caused by a defect in the mitochondria.^[9-11] Oncocytic cells are most commonly found in the salivary glands, thyroid, parathyroid, and kidney, with the parotid gland being the most common site.^[8,11] Oncocytic tumors of salivary glands included in the 2005 WHO histologic classification are oncocytoma, Warthin tumor and oncocytic carcinoma.^[2]

Oncocytic lipoadenoma was first described by Hirokawa et al.^[3] in the submandibular gland in 1998. The clinical and pathologic features of the cases reported in the English literature are summarized in Table 1.^[3-6] Three patients were female; two were male. Four involved the parotid gland; one was located in the submandibular gland (Table 1). The masses described in the literature were painless, soft, well-defined, mobile and slow-growing.^[3,5,6] The duration of symptoms ranged between three months and 10 years.^[3,5,6] The current case presented with a two-year history of a painless, slow-growing mass of the parotid gland. Imaging findings of oncocytic lipoadenoma may be variable. The findings of cervical ultrasonography and magnetic resonance imaging supported a diagnosis of vascular tumor in one of the cases.^[6] Computed tomography scan showed an ill-defined mass with "lipoma-like" areas in another case.^[5] Computed tomography findings suggested a diagnosis of pleomorphic adenoma in our case. Fineneedle aspiration specimens, described in three cases including the current case, showed oncocytic cells.^[5,6]

Oncocytic lipoadenoma is an encapsulated neoplasm composed of oncocytic cells and mature adipose tissue. It is composed of oncocytes similar to those of conventional salivary gland oncocytoma with "light" and "dark" cells.^[8,10] It may reveal ductal components. Klieb and Perez-Ordoñez^[5] first described sebaceous differentiation in oncocytic lipoadenoma. This is the second case with sebaceous glands surrounded by small lymphocytes.

The differential diagnosis of oncocytic lipoadenoma includes nodular and diffuse oncocytic hyperplasia, oncocytoma, and sialolipoma. Nodular oncocytic hyperplasia is nodular accumulation and proliferation of oncocytes in the salivary glands, and is usually multiple and not encapsulated. In diffuse oncocytic hyperplasia, almost all of the salivary gland manifests oncocytic metaplasia, and may be associated with fatty infitration. However, it does not form an encapsulated mass.^[3] Oncocytoma has a fibrous capsule, but the nodule is composed entirely of oncocytic cells.^[12] Sialolipoma is a well-delineated and/or encapsulated neoplasm composed of mature adipose tissue containing foci of atrophic, non-neoplastic, salivary gland acini and ducts, entrapped within the lipomatous mass. The lipoadenoma is composed of neoplastic epithelial ductal components in addition to neoplastic adipose tissue.^[13]

A study of the immunohistochemical profile of oncocytic lipoadenoma demonstrated that oncocytes were positive for low-molecular-weight keratin, keratin 7, and epithelial membrane antigen, with only a small subgroup of cells expressing high-molecular-weight keratin, keratin 5/6, keratin 19, and p63. The epithelial component of oncocytic lipoadenoma lacked staining for actins and calponin. These findings suggest the presence of dual epithelial population with ductal and partial basal-cell differentiation and the absence of a myoepithelial participation within the tumor.^[5]

Oncocytic lipoadenoma is a rare entity. It should be considered in the differential diagnosis of other oncocytic proliferations and oncocytic tumors in the parotid gland.

REFERENCES

- 1. Batsakis JG. Tumors of the head and neck. Clinical and pathological considerations. 2nd ed. Baltimore: Williams & Wilkins; 1979.
- Tumors of the salivary glands. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization Classification of Tumors: Pathology and genetics of tumors of head and neck. Lyon: IARC Press; 2005. p. 209-81.
- Hirokawa M, Shimizu M, Manabe T, Ito J, Ogawa S. Oncocytic lipoadenoma of the submandibular gland. Hum Pathol 1998;29:410-2.
- 4. Kato M, Horie Y. Oncocytic lipoadenoma of the parotid gland. Histopathology 2000;36:285-6.
- Klieb HB, Perez-Ordoñez B. Oncocytic lipoadenoma of the parotid gland with sebaceous differentiation. Study of its keratin profile. Virchows Arch 2006; 449:722-5.
- Aouad R, Matar N, Sader-Ghorra C, Haddad A. Pathology quiz case 1. Oncocytic lipoadenoma of the parotid gland. Arch Otolaryngol Head Neck Surg 2008;134:446-8.

- Kandiloros D, Segas J, Papadimitriou K, Koutsomanis P, Adamopoulos G. Malignant oncocytoma of the parotid with oncocytic change of the contralateral gland. Am J Otolaryngol 1995;16:200-4.
- 8. Ellis GL, Auclair PL. Benign epithelial neoplasms. In: Rosai J, Sobin LH, editors. Tumors of the salivary glands. Atlas of Tumor Pathology. 3rd ed. Washington: Armed Forces Institute of Pathology; 1996. p. 39-153.
- 9. Takeda Y. Diffuse hyperplastic oncocytosis of the parotid gland. Int J Oral Maxillofac Surg 1986;15:765-8.
- 10. Brandwein MS, Huvos AG. Oncocytic tumors of major salivary glands. A study of 68 cases with follow-up of 44 patients. Am J Surg Pathol 1991;15:514-28.
- Chang A, Harawi SJ. Oncocytes, oncocytosis, and oncocytic tumors. Pathol Annu 1992;27:263-304.
- Palmer TJ, Gleeson MJ, Eveson JW, Cawson RA. Oncocytic adenomas and oncocytic hyperplasia of salivary glands: a clinicopathological study of 26 cases. Histopathology 1990;16:487-93.
- Ramer N, Lumerman HS, Ramer Y. Sialolipoma: report of two cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007; 104:809-13.