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Oncocytic sialolipoma, a rare entity: a case report with review of literature

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ABSTRACT

Sialolipoma is a rare benign neoplasm of salivary gland composed of neoplastic mature adipose tissue and non-neoplastic salivary gland elements. Oncocytic sialolipoma is a very rare tumor which is considered to be a variant of sialolipoma and it harbours nodules of oncocytic salivary glandular tissue admixed with lipomatous areas. Even though 51 cases of sialolipoma were reported so far only 3 cases of them showed features of oncocytic sialolipoma. Here we present fourth case of oncocytic sialolipoma in a 49 year old male who presented with parotid swelling of three years duration. FNAC done outside was suggestive of pleomorphic adenoma. Superficial parotidectomy was done and it was diagnosed as oncocytic sialolipoma.

Keywords: Sialolipoma, Oncocytic, Parotid

INTRODUCTION

Sialolipoma is a benign rare neoplasm of salivary gland. Extensive literature search showed only 51 cases of sialolipoma of the salivary gland reported worldwide.[1] Clinical features are similar to pleomorphic adenoma with very less incidence of facial nerve entrapment (14%).[1,2]Incidence is more common in males. The common age of presentation is fifth to sixth decade. Even though numerous histological variants of salivary gland lipoma have been described oncocytic sialolipoma is considered to be a rare variant and only 3 cases are reported so far.[2,3,4,] Hence we are reporting fourth case of oncocytic sialolipoma in a 49-year old male along with review of previous three cases.

CASE REPORT

A 49 year old man, smoker, presented with left parotid swelling of 3 years duration. It was a slow growing painless round mass with soft to doughy consistency .An ultrasonogram showed 2.7 cm x 1.2 cm sized normal echogenic nodule with well-defined margins suggestive of parotid adenoma. FNAC done outside was suggestive of pleomorphic adenoma. Superficial parotidectomy was done and we received slides and blocks for review .Grossly it was a well circumscribed nodular mass measuring 2.5x2.5x1cm.Cut section was yellowish with faint lobulations.

Microscopy showed a neoplasm composed of nodules of oncocytic cells with densely eosinophilic cytoplasm and uniform small nuclei admixed with sheets and lobules of mature adipocytes and islands of salivary gland acini with ducts in between.These were seen scattered among the adipose tissue. Fat constituted about 60% of the tumour. Periductal lymphocytic infiltrate was noted(Fig. 1,2).The salivary ducts in between showed sebaceous metaplasia (Fig .3) . A thin rim of fibrous capsule was noted in the periphery separating it from normal salivary gland tissue.(Fig.4)

DISCUSSION

Sialolipoma is a newly recognised variant of lipoma showing proliferation of benign salivary gland tissue intimately admixed with mature adipose tissue. Nagao et al in 2001 reported the first case of sialolipoma.[5] This entity belongs to a family of fat-containing tumors of the salivary glands including lipoma,non-oncocyticsialolipoma,oncocytic

lipoadenoma and pleomorphic adenoma/myoepithelioma with extensive lipometaplasia. The WHO 4 th edition head and neck tumors describes sialolipomas under the soft tissue tumors of the salivary glands as "lipomas entrapping salivary glandular tissue," but not as mixed tumors. WHO describes sialolipoma and oncocytic lipoadenoma synonymically(6) Similar salivary gland tumors exhibiting features of both oncocytic lipoadenoma and sialolipoma have been illustrated by other investigators, one of which was described using the term "oncocytic sialolipoma".[2,3,4] Whether to classify such lesions as oncocytic lipoadenoma, oncocytic sialolipoma, or as a hybrid lipoepithelial salivary gland tumor remains to be determined. Nonetheless, the existence of these phenotypically mixed cases suggests a possible histogenetic relationship between oncocytic lipoadenoma and sialolipoma. Presence of histological findings like lobules of lipocytes, sebaceous metaplasia, periductal chronic inflammation and periductal fibrosis, overlap with those of oncocytic lipoadenoma which may suggest an association between these entities.(7) Immunohistochemical findings in the form of cytokeratin profile, P63 and cytogenetic studies carried out so far also suggest that there is significant association between oncocytic lipoadenoma and sialolipoma(7).The ducts and acini in oncocytic sialolipoma are shown to have normal cellular phenotype both by immunohistochemistry and electron microscopy. These findings suggest that normal salivary gland tissue is entrapped in the mass during adipocytic proliferation rather than representing neoplastic elements.(8) Regarding the histogenesis of sialolipoma Akrish et al suggested dysfunction of salivary gland.(9) The factors favoring this theory include the long duration, atrophic acinar and ductal components, fibrosis,dialated salivary ducts, oncocytic metaplasia and squamous metaplasia. In our case the patient had swelling for three years supporting the long duration in the above theory.

.Oncocytic sialolipoma, rare variant of sialolipoma was first reported by Pusiol et al in 2009 in submandibular gland. (2) The formation of oncocytic micronodules has been considered as a result of oncocytic metaplasia and hyperplasia of small and large ducts. [10] In the first case, oncocytic nodules were adjacent to ductal structures and isolated in the fatty tissue.It was in a 73yr old male with fatty tissue constituting about 80% and it showed sebaceous differentiation. The second case was in 2014 in a 43 yr old female again in submandibular gland by Ahn D et al.(3)The fat constituted about 80% of the tumour and there was no sebaceous differentiation. Ruangritchankul et al reported the third case of oncocytic sialolipoma in the parotid gland in a 73 year old female.(4) Adipose tissue component was 85% of the total volume and there was no sebaceous differentiation. This is the fourth case which is in a 49 year old male with parotid swelling. 60% of the tumor was composed of mature adipose tissue which agrees with the literature review of sialolipomas, all of which says adipocytic component predominates in them.(1) The fat constitutes about 90% in major salivary glands compared to minor salivary glands. Sebaceous differentiation was reported more in oncocytic lipoadenoma compared to sialolipoma. However our case and first case by Pusiol et al showed sebaceous differentiation. As in other 3 cases of oncocytic sialolipoma there were multiple oncocytic nodules with dialated ducts and periductal lymphocytic collections. Our patient was a smoker even though no relation to smoking was established in oncocytic lipoadenoma or oncocytic sialolipoma. The presence of normal salivary gland with duct dialatation and fibrosis precludes the possibility of a pleomorphic adenoma with extensive adipose tissue content. If a tumor has oncocytic nodules as in our case, it should be differentiated from oncocytoma, which has some malignant potential. Distinguishing between these 2 tumor types is not difficult because oncocytic nodules of sialolipoma are surrounded by abundant adipocytes adjacent to normal glandular structures while in oncocytoma there are no adipocytic lobules. The fibrous capsule in these cases ruled out nodular oncocytic metaplasia which is an unencapsulated multifocal proliferation of oncocytes and is a close differential for oncocytic sialolipoma.. Ruangritchankul et al says his case is the fourth case of oncocytic sialolipoma and third case was reported by Agaimy et al in his series of 31 cases of lipomatous salivary gland tumors.(11)But in his article Agaimy describes four cases of non oncocytic sialolipomas only. Not even a single case of oncocytic sialolipoma was mentioned.So Ruangritchankul described third case of oncocytic sialolipoma in 2018 following Pusiol T et al in 2009

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and Ahn D et al in 2014.The present case is the fourth case of oncocytic sialolipoma

Conclusion

Oncocytic Sialolipoma is a very rare, but distinct salivary gland neoplasm. It could develop in any salivary gland .As a newly described tumor type, care should be taken to distinguish it from other salivary gland neoplasms such as simple lipoma, pleomorphic adenoma, or oncocytoma. Sialolipoma and lipoadenoma with or without oncocytosis and/or sebaceous differentiation should be considered organ-specific tumors with a distinct histological appearance and specific terminology.

Conflict of Interest:NIL

References

1. Khera S, Elhence P, Bagga N, Garg P, Choudhary R. Sialolipoma of Parotid Gland: a Lesser Known Entity. Indian Journal of Surgery. 2020 Feb;82(1):90-2.
2. Pusiol T, Franceschetti I, Scialpi M, Piscioli I. Oncocyticsialolipoma of the submandibular gland with sebaceous differentiation: A new pathological entity. Indian J PatholMicrobiol 2009;52:379-82
3. Ahn D, Park TI, Park J, Heo SJ. Oncocyticsialolipoma of the submandibular gland. Clinical and experimental otorhinolaryngology. 2014 Jun;7(2):149.
4. Ruangritchankul K, Connor S, Oakley R. OncocyticSialolipoma of Parotid Gland: Case Report and Literature Review. Head and neck pathology. 2018 Dec 11:1
5. Nagao T, Sugano I, Ishida Y, Asoh A, Munakata S, Yamazaki K, et al . Sialolipoma: A report of seven cases of a new variant of salivary gland lipoma. Histopathology 2001;38:30-6
6. El-Naggar AK, Chan JK, Grandis JR, Takata T, Slootweg PJ, editors. WHO classification of head and neck tumours. International Agency for Research on Cancer; 2017.
7. Lau SK, Thompson LD. Oncocyticlipoadenoma of the salivary gland: a clinicopathologic analysis of 7 cases and review of the literature. Head Neck Pathol. 2015 Mar; 9(1): 39–46.
8. Ilie M, Hofman V, Pedeutour F, et al. Oncocyticlipoadenoma of the parotid gland: Immunohistochemical and cytogenetic analysis. Pathol Res Pract. 2010;206:66–72.)
9. Akrish S, Leiser Y, Shamira D, Peled M. Sialolipoma of the salivary gland: Two new cases, literature review, and histogenetic hypothesis. J Oral Maxillofac Surg. 2011;69:1380–4
10. Brandwein MS, Huvos AG. Oncocytic tumors of major salivary glands. A study of 68 cases with follow-up of 44 patients. Am J SurgPathol 1991;15:514-28
11. Agaimy A, Ihrler S, Märkl B, Lell M, Zenk J, Hartmann A, Michal M, Skalova A. Lipomatous salivary gland tumors: a series of 31 cases spanning their morphologic spectrum with emphasis on sialolipoma and oncocyticlipoadenoma. Am J SurgPathol. 2013;37:128–137. do.4103/0377-4929.55000

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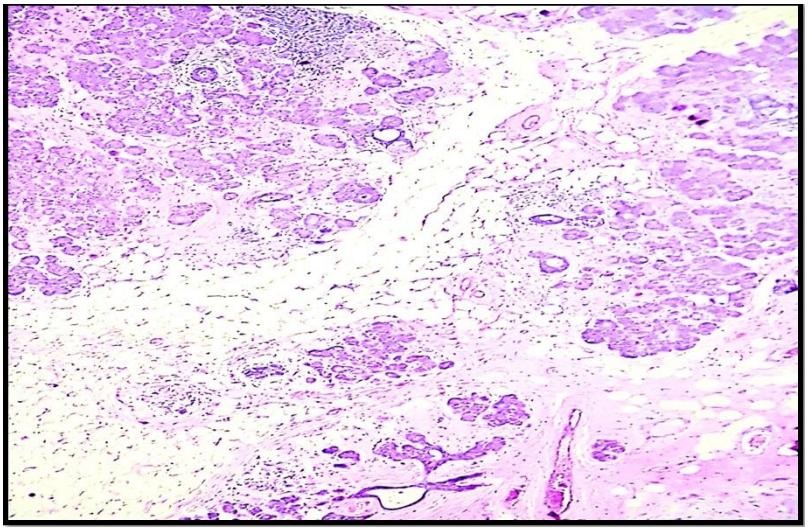

Fig1

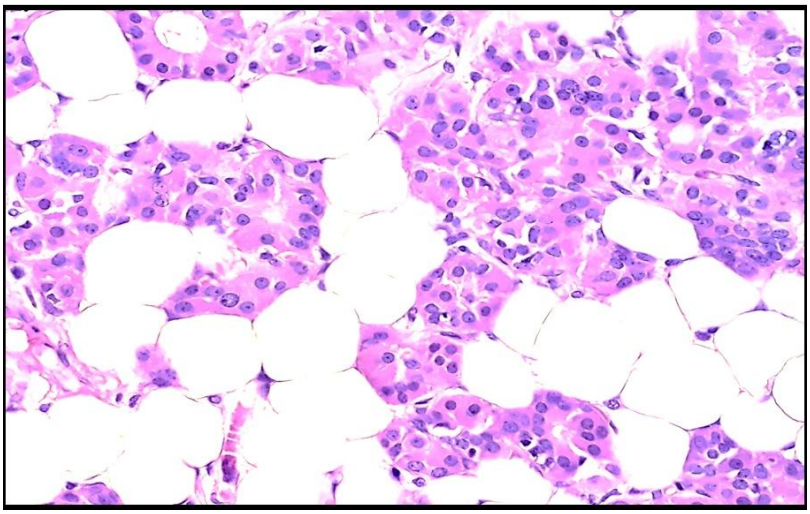

Fig 2

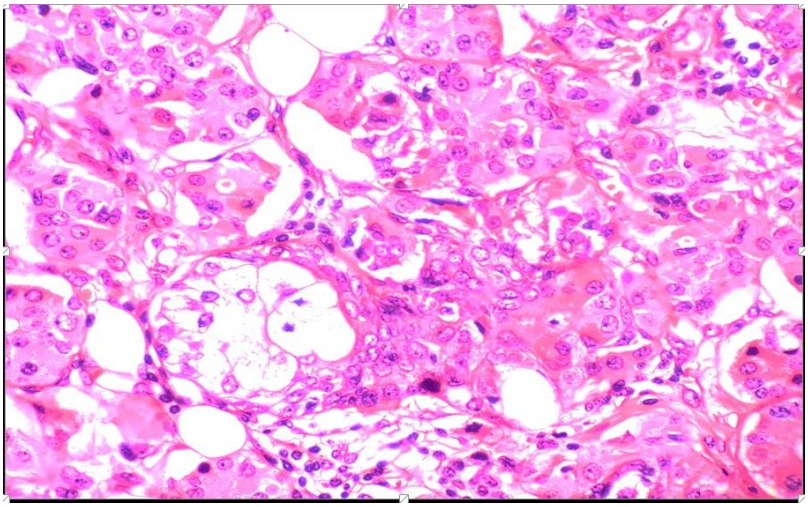

Fig 3

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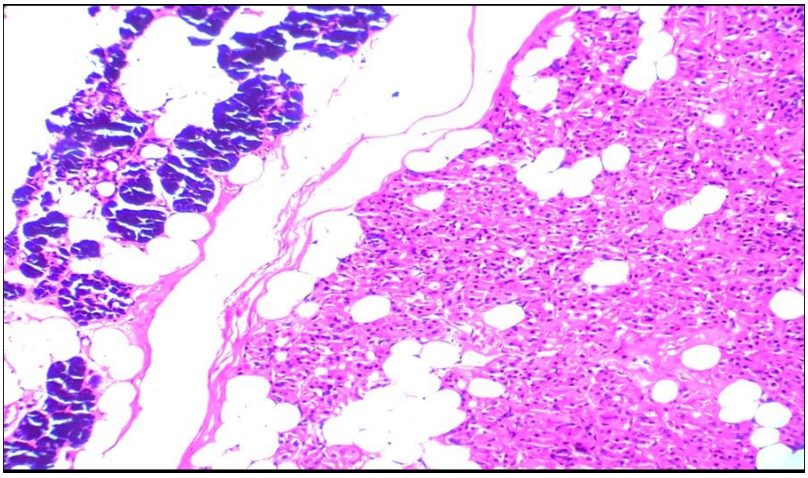

Fig 4

Fig.1.*40x showing oncocytic nodules with adipose tissue and entrapped salivary gland tissue in between.*

Fig.2.H&E 200x showing oncocytic nodules admixed with adipose tissue.

Fig.3. H&E 400 x showing salivary duct with sebaceous metaplasia.

Fig.4. H&E 100x showing normal salivary gland and well circumscribed tumor with a thin rim of fibrous capsule in the periphery.

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