Solitary Oncocytoma of the Submandibular Salivary Gland in an Adolescent Female : A Case Report

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Abstract

We report a rare presentation of Oncocytoma of the submandibular salivary gland in an young adolescent female who presented with progressive swelling in the left submandibular region since childhood with pain for the past one month. Fine needle aspiration revealed Oncocytoma for which a conventional submandibular excision was done. There are few articles about the rare presentation of submandibular Oncocytoma and literature favors older population for this rarity. This case was unique because of its presentation in a young individual and there were no supporting published articles available till date.

Keywords Salivary gland tumors · Submandibular gland neoplasm · Oncocytoma · Oxyphil adenoma

Introduction

Oncocytomas are rare tumors that constitute only 2.3% of benign epithelial salivary gland neoplasms. They are most often encountered after the sixth decade of life with a nearly equal male to female ratio of occurrence⁹. The majority of these tumors affect the parotid gland (78%), few affect the submandibular gland (9%). The clinical presentation of Oncocytomas is essentially identical to other benign salivary tumors – a slow growing, non tender mass². They are firm, may be multilobulated and mobile on examination. Oncocytomas, along with Warthin's tumor have been noted to demonstrate increased uptake of pertechnetate anion and

therefore can be distinguished from some other neoplasm by using technetium-99m pertechnetate scintigraphy^{3,4}. Standard treatment of oncocytoma is surgical excision with a margin of normal tissue. There is an extremely low rate of recurrence of these tumors if removal is complete. Enucleation or curettage is not appropriate¹¹.

Case Report

A 19-year-old normotensive euglycemic female presented with swelling in the left side of the neck below the mandible since childhood. (Fig.1) It was insidious in onset and was small in size to start with and progressive in nature. To start with it was painless but as she grew older she experienced dull aching pain which was more pronounced over the past one month prior to hospitalization. Swelling was not associated with difficulty in swallowing nor was there an increase in size during mastication. She did not give any history of dryness of mouth or fever.

Examination at our clinic revealed a bimanually palpable asymptomatic swelling measuring 3 cm X 5 cm in size in the left submandibular region. Swelling was tender, firm in consistency with healthy skin covering it. There was no local rise of temperature though. No nodal involvement was made out. Examination of the ENT and other systems were found to be normal. CT scan neck revealed an encapsulated left submandibular swelling (Fig.2).

Fine needle aspiration cytology revealed tumor cells filled with mitochondria, which were easily demonstrated by histochemical reactions suggestive of epithelial adenoma or Oncocytoma (Fig.3). Under General Anesthesia the left submandibular gland excision was done by the conventional method and sent for Histopathological examination (Fig.4). Post operative period was uneventful. The patient was treated with antibiotics and analgesics for five days. There was no clinical evidence of neurological deficit and was discharged with good health on day five after suture removal. Patient was comfortable at review after 8 weeks.

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Fig. 1 Swelling in the left submandibular region.

Fig. 2 CT scan axial section of the neck showing the left submandibular salivary gland swelling.

Fig. 3
Histopathological picture of Submandibular salivary gland oncocytoma X 400 Showing nests of oncocytes with acidophilic granular cytoplasm (H&E stain)

Fig. 4 Oncocytoma – Gross appearance.

Discussion

Oncocytomas are rare benign tumors epithelial salivary gland neoplasms¹. They are most often encountered after the sixth decade of life with equal male to female ratio of occurrence⁵. It constitutes 9 % of all submandibular gland neoplasms with peak occurrence in the sixth and seventh decade¹⁰. First reported case of oncocytoma was dated back to 1965 by Enroth et al¹². The largest series of 22 cases reported the disease only in older individuals with one individual at the age of 21¹⁰. Brandwein et al series of 68 cases of major salivary oncocytoma which included 14 cases of sub-mandibular origin involved patients with age group between 38 and 70 years^{6,7,8}.

These were slow growing asymptomatic mass but occasionally associated with pain which was comparable with our case report¹⁰. We had a working diagnosis of pleomorphic adenoma considering the age and presentation. To our surprise it was reported to be an oncocytoma of the submandibular gland. There was only one supporting article published so far regarding oncocytoma in a adolescent age group¹⁰.

After diagnosis, submandibular gland excision was done in the conventional method through sub-mandibular incision². There were articles supporting radiotherapy as an alternative treatment with reported increased rates of recurrence⁸.

This report was published to emphasize the need for a histopathological proof to support the final tissue diagnosis

and odd presentations of tumors especially those which are rare, like oncocytoma of the submandibular gland.

Conclusion

This case highlights the need for increased awareness among pathologists, otolaryngologists and Head & Neck surgeons about the possibility of Oncocytomas arising in the Submandibular Gland in the second and third decade of life. The complete excision of the Submandibular Gland is the treatment of choice with an extremely low rate of recurrence.

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