Osteoclast- like Giant Cell Tumour of the Salivary Gland: Case Report and Literature Review

P Brown[1], J Mamby-Alexander[2], G Channer[1]

ABSTRACT

The Osteoclast- like Giant cell tumour of the salivary gland is very rare and 24 cases have been reported so far in the English literature. There is no published literature in the West Indies regarding this rare lesion. This case report aims to highlight an additional case emphasizing pathology, management and a recent literature review.

Keywords: Giant cell tumours parotid, parotid neoplasm, salivary gland tumours

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From: [1]Department of Surgery, Ear, Nose and Throat division, Kingston Public Hospital, Kingston, Jamaica. [2]Surgipath and Cytology Services, Kingston, Jamaica.

Correspondence: Dr P Brown, Department of Surgery, Ear, Nose and Throat division, Kingston Public Hospital, North Street, Kingston, Jamaica. E-mail: drphillipbrown.ent@gmail.com

INTRODUCTION

The Osteoclast like Giant cell tumour of the salivary glands(OGCT-SG) is a very rare salivary gland pathology with a poorly understood histiogenesis (1). It was initially described by Eusebi et al in 1984(2). Since then, only 24 cases have been reported so far in the English literature, to the best of our knowledge(3, 4). These cases have varied demographics, pathologies, treatment approaches and outcomes. We aim to report the 25th case, highlighting the clinicopathological features, management approaches and the latest literature review. This is the first reported case from Jamaica and the Caribbean.

CASE REPORT

A 44 year old female presented with a one year history of a right sided, slowly growing, asymptomatic parotid mass. This mass was 3x2cm, firm, non-tender and located in the preauricular region. There was no associated lymphadenopathy and the facial nerve was normal. She had an Ultrasound (US) and Computer Tomography (CT) which confirmed a lobulated firm parotid mass. Her Ultrasound guided Fine needle aspiration cytology (US-FNAC) demonstrated a poorly differentiated carcinoma with spindle cell features and giant cell formations (Figure 1). She had a right subtotal parotidectomy with no significant postoperative sequelae. The final histopathology demonstrated an Osteoclast- like Giant Cell Tumour of the parotid gland without a carcinomatous component (Figure 2). She has no evidence of recurrence up to 3 years follow up.

DISCUSSION

Primary Giant cell tumours of the salivary gland are very rare with only 24 previous cases described(3, 4). This report highlights the 25th case. Giant cell tumours classically affect bone but have been reported to affect soft tissue and visceral organs (3, 5, 6). These tumours are described as osteoclast type or osteoclast type giant cell due to similarities to giant cell tumour of bone morphologically, histologically and clinically (3).

A recent literature review of all cases (3,4,7-9) demonstrated that OGCT-SG primarily affects the parotid gland (83%) followed by the submandibular gland (12.5%) and minor salivary gland (4.5%). This finding is consistent with the index case. The common presentations albeit variable include rapid growing mass with/without pain and no facial nerve involvement(9). Metastatic lymphadenopathy has been described in two cases (1, 9). It affects men more with a male to female ratio of 4:1. The average age of the patients was 58 years old ranging from 28 to 92 years old. These demographics were not typical of our index case which typifies the wide clinical spectrum of OGCT-SG.

OGCT-SG can occur with or without a carcinomatous component. The previous reported cases (3, 4, 9) demonstrated that carcinoma occurred in 62.5% (15/24). These include salivary duct carcinoma in the majority of cases and carcinoma ex pleomorphic adenoma. It is therefore prudent that specimens be properly examined with a diagnosis of OGCT-SG to rule out the presence of a carcinoma. Carcinomas are associated with increased metastatic risk (1, 9) and therefore subtends poorer prognosis. Our index case tumour after extensive histomorphological examination had no evidence of a carcinoma. The literature review demonstrated that nine patients (3, 4, 7, 9, 10) were followed for greater than 9 months. Seven are alive without

evidence of disease whilst two died from pulmonary metastatic disease. Our index case at 3 years of follow up, longest reported to date, had no evidence of disease or recurrence.

OGCT-SG despite the similar histomorphological features to Giant cell tumour of bone has some contrasting characteristics. These include OGCT-SG strong association with carcinoma (62.5%) and its biological aggressiveness. In addition, OGCT-SG lacks peripheral reactive bone formation (1,3,7, 9). Furthermore, Giant cell tumour of the mandible or maxilla with parotid gland invasion must be ruled out clinicoradiologically or intraoperatively. This was excluded in our index case.

The exact histiogenesis and tumour classification of OGCT-ST have been undefined and controversial in the literature. Some authors (2,3,7,11) believe that OGCT-ST is a carcinoma and prefer the term osteoclast like giant cell carcinoma. These are based on the strong association with carcinoma (62.5%), mononuclear cells expression of both epithelial and histiocytic markers(1), similar allelic mutation 17p13 as salivary duct carcinoma(9) and the microsatellite pattern of the giant cell component resembling a carcinoma(3). It is thought that the osteoclast like giant cells are likely reactive and a non-neoplastic component as they lack epithelial markers and possess CD68 histiocytic marker (1, 11). The World Health Organization (WHO) ascribes OGCT-ST as an undifferentiated carcinoma without any distinct categories. Some authors (1, 3, 11) disagree with this notion based on the male predominance and more favourable prognosis of OGCT-SG compared to undifferentiated carcinoma.

This controversy even extends to the definition of optimal surgical management. The diagnostic accuracy of FNAC, extent of parotidectomy, addressing the facial nerve, role of neck dissection and role of radiotherapy are all controversial areas that is ill defined in the literature. The optimal approach has to be individualized based on patient, pathological, surgical and

institutional factors and preferences. The key principle is total surgical excision with minimal morbidity and to rule out an associated carcinoma as was done in the index case.

CONCLUSION

OGCT-ST is a rare salivary gland tumour whose histiogenesis and surgical management remains ill defined. It has a strong male predominance, varied age range and clinical presentation. It has a strong association with carcinoma (62.5%) and therefore it is mandatory to rule out this in tumour specimens. Its presence portends a higher risk of metastasis and worse prognosis. The optimal management has to be individualized. We present the 25th case, first in Jamaica and Caribbean, in a 44 year old female with the longest follow up to date (3yrs) who has no evidence of disease or recurrence.

AUTHORS' NOTE

P Brown conceived the paper, wrote the manuscript and approved the final version. G Channer edited and approved the final version. J Mamby-Alexander was instrumental in the provision of the pathology slides and approval of the final version.

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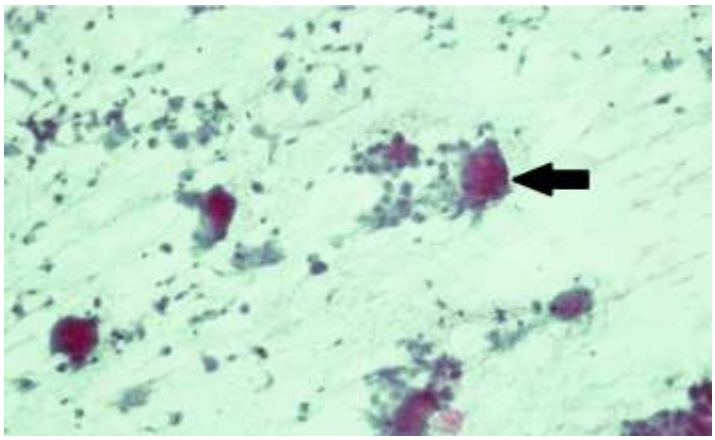

Fig. 1: FNAC demonstrating poorly differentiated carcinoma with spindle cell features and giant cell formation (bold arrow).

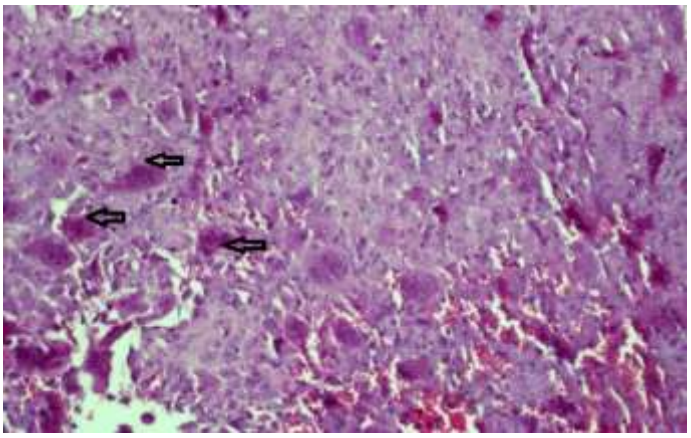

Fig. 2: Final histology demonstrating an Osteoclast-like Giant cell tumour of the parotid gland (open arrows-Giant cell formations).