

Malignant Myoepithelioma of the Parotid Gland: Literature Review on the Occasion of Our Report of a Case

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ABSTRACT

Malignant myoepitheliomas are rare malignant salivary gland neoplasms which usually occur in the parotid gland. Very little material on this topic has been published. This tumour represents a therapeutic challenge since there are no specific recommendations or guidelines. We present a case of malignant myoepithelioma arising de novo in the parotid gland. We discuss our therapeutic approach and considerations on appropriate post-surgical treatment. Then we provide a review of international literature on this interesting and rare oncological entity.

Keywords: Malignant Myoepithelioma; Parotid; Radiotherapy; Rare Neoplasms; Review

1. Introduction

Myoepitheliomas of the salivary glands are rare tumours, accounting for about 1% of all salivary gland tumours [1-3]. Most of them are benign and only 10% are malignant [1]. Most malignant myoepitheliomas occur in the parotid gland [1,4,5].

To our knowledge, in medical publications in English, the first report of a case of a malignant myoepithelioma (MM) of a minor salivary gland was in 1975 by Stromeyer *et al.* [6] while the first case of a MM of the parotid gland was reported in 1977 by Crissman *et al.*; the latter presented the case of a locally aggressive MM that eventually metastasized [7]. Further reports on malignant myoepitheliomas (MMs) have been generally scarce.

Our search in pubmed, particularly focused on the last 35 years (between the years 1975 and 2010) revealed only a few dozen posts on salivary gland myoepitheliomas. Most of these concerned tumours of the parotid gland, whilst a smaller number of them were about minor salivary gland tumours. There have also been sporadic reports of MM at unusual sites, e.g., the breast, bone, external auditory canal, skin and vulva. The majority of these reports deal with the clinical and pathologic or the immunohistochemical features and complexities of the tumour [4,8-18] but offer limited data concerning the treatment.

In this paper, written on the occasion of a patient presenting with an MM of the parotid, we provide a review of previous publications concerning the issue.

2. Case Report

A 72-year old man presented with a painless mass in the right parotid region. The mass, which had progressed slowly the last few months, was firm on examination. The patient was referred for a fine needle aspiration and the cytology revealed a malignant epithelial tumour.

The patient was submitted to a partial right parotidectomy with free margins and lymph node dissection. The histological analysis showed a grade 2—malignant myoepithelioma predominantly made up of epithelioid cells but a spindle cell component was also present. Mitotic figures and nuclear pleomorphism were also observed. Neoplastic cells were growing as multiple nodules with extended central necrosis. These results match the description of an MM [11]. There were focal regions of tumour cells infiltrating the capsule but tumour infiltration into the adjacent parotid tissue was absent. Immunohistochemical reaction was strongly positive for S-100 protein and vimentin.

The patient was referred for postoperative radiotherapy. Using two ipsilateral 6 MV photon beams [anterior oblique and posterior oblique] we delivered a total dose of 60 Gy to the tumour bed and the right parotid region, with a conventional fractionation of 2 Gy/fraction, five fractions per week, for a total duration of six weeks. A thermoplastic mask was used as an immobilization device during the simulations and the treatment as a whole.

The patient completed the irradiation with a satisfactory treatment tolerance; acute toxicity included only

grade I-II dermatitis and a slight, grade I-toxicity of the buccal mucosa was evident. Eleven months after the completion of the radiotherapy treatment the patient died as a result of undercurrent heart disease, without any evidence of local or regional relapse of the myoepithelioma or any late side effect of the radiotherapy.

3. Review

Myoepitheliomas of the salivary glands are rare tumours that occur mostly in the parotid gland [1,4,5]. However, other head and neck sites of origin such as the submandibular gland [19,20] oral cavity [21], palate [3,4,22-25] larynx [26,27], parapharyngeal space [28], nasopharynx [29-32], maxillary sinus [33], external auditory canal [34] have, rarely, been reported in the literature. MMs have also been, sporadically, reported in unusual locations, e.g., the breast [35-37], bone [38-40] skin [41] and vulva [42].

Malignant myoepithelioma of the parotid may occur within pre-existing benign lesions like pleomorphic adenomas [8,9,13,43-45] or benign myoepitheliomas, but may also occur de novo [9,13,19,43,46,47]. These tumours are distinguished from their benign myoepithelial counterparts by their infiltrating and destructive growth, cellular pleomorphism and mitotic figures [48]. According to a recent report by Darvishian and Lin the malignant features of myoepithelial carcinoma arising mainly in the salivary glands are pleomorphism, coarse chromatin, prominent nucleoli, mitotic figures and/or necrosis [49].

Histologically, MMs are composed of one or several cell types: spindle, plasmacytoid, epithelioid and clear cells [23]. The mean age of presentation is 62 years [1] and a slow-growing painless mass is usually involved [28].

Imaging features of myoepitheliomas of the parotid gland rarely appear in the literature because of the low prevalence of the tumours. By means of ultrasonography, CT and MRI, myoepitheliomas have been described to be

well circumscribed, mass lesions with smooth or lobulated margins. They may be solid lesions or contain both cystic and solid components. Following intravenous contrast medium injection at CT and MRI, the lesions may exhibit either homogeneous or inhomogeneous enhancement. They are usually depicted in the superficial lobe of the parotid gland and less frequently found in the deep lobe or the tail of the gland [50-52].

As myoepithelioma of the parotid is not a common tumour, there are no specific guidelines for its treatment. Complete resection with wide margins is considered essential and has given favourable outcomes for salivary gland myoepitheliomas [5,14,22]. Until now the mainstay of treatment has been surgery [22,28]. Malignant myoepithelial cell-rich lesions require a wider local excision and lymph node dissection [49].

According to a retrospective study of 27 cases of myoepithelial carcinoma of the salivary glands the tumour is characterised by extensive local growth and invasion of the surrounding tissues. Although cervical lymph node metastasis is infrequent [53], there are high rates of distant metastases [5].

There are reports of malignant myoepitheliomas presenting with secondary lesions to the lung [10,54] liver [26], skin [55] or lymph nodes [10], while others have a tendency to recur locally [10,54].

For our review, some of the above data are summarized on the tables, *i.e.*: **Table 1** summarizes the references of MMs of salivary glands (all sites), and **Table 2** summarizes the number of MMs of parotid gland referred in the international literature with the special features of each reference.

There is extremely limited data concerning radiotherapy for this rare tumour. The first use of radical radiotherapy in MM was reported in 2009, concerning a 90-year-old man with impaired performance status and a MM of the soft palate. Radiotherapy gave excellent palliative effect and local control [22].

In our case, presented in this paper, the patient was re-

Table 1. Malignant myoepitheliomas of salivary glands: number of cases per reference and per site.

References	Total cases	Cases per site			
		Parotid	Submandibular	Sublingual	Minor salivary glands
Nagao <i>et al.</i> (1998) [1]	10	7	2	---	1 (palate)
Sciubba <i>et al.</i> (1982) [2]	23	12	5	---	6 (5 intraoral, 1 upper lip)
Yu G. <i>et al.</i> (2003) [5]	27	14	4	1	8 (7 palate, 1 base of tongue)
Savera <i>et al.</i> (2000) [47]	25 ^a	15	4	---	5 (2 palate, 1 maxill sinus, 1 tongue (base), 1 supragl larynx)
Di Palma <i>et al.</i> (1993) [54]	10	8	---	---	2 (1 palate, 1 cheek)
Alos <i>et al.</i> (1996) [55]	4 ^b	2	---	---	2 (NOS)

^acase with cervical lymph node metastases of unknown primary site; ^bthey had twelve benign and four malignant myoepitheliomas.

Table 2. Malignant myoepitheliomas of the parotid gland: number of cases per reference and special features.

MM of parotid gland	No of cases	Special features
Crissman <i>et al.</i> (1997) [7]	1	PMM locally aggressive and eventually metastasized
DiPalma <i>et al.</i> (1996) [8]	5	Almost all PMMs initially thought to be recurrent pleomorphic adenomas
Singh <i>et al.</i> (1988) [9]	1	PMM arising in a pleomorphic adenoma
Klumpp <i>et al.</i> (1995) [10]	1	Local, regional and metastatic progression prior to completion of adjuvant RT
Chow <i>et al.</i> (1996) [11]	1	PMM's cytologic features in FNA
Sironi <i>et al.</i> (1997) [12]	1	PMM's cytologic, immunocytochemical and ultrastructural study
Daneshbod <i>et al.</i> (2007) [13]	1	PMM arising in a pleomorphic adenoma
Gun <i>et al.</i> (2009) [15] ¹⁵	1	PMM with histological uncertainties
Siddarayu <i>et al.</i> (2008) [16]	1	PPM's preoperative FNA cytologic diagnosis
Shinozaki <i>et al.</i> (2008) [17]	42	Six out of forty-two with sebaceous differentiation (histological variant of PMM)
Lauro <i>et al.</i> (2003) [43]	1	PMM arising in a pleomorphic adenoma
Di Palma <i>et al.</i> (1991) [44]	1	PMM arising in a pleomorphic adenoma
Di Palma <i>et al.</i> (1998) [45]	1	PMM arising in a pleomorphic adenoma
Dardick <i>et al.</i> (1989) [7]	5	PMM's histological, ultrastructural, and immunocytological features
Maurer <i>et al.</i> (2009) [50]	1	CT and MRI in the differentiation between PBM and PMM
Wang <i>et al.</i> (2008) [51]	10	PMM's CT imaging findings
Lee <i>et al.</i> (2005) [52]	2	PMMs clinically suspected to be infra-auricular subcutaneous epidermal cysts

MM: Malignant Myoepithelioma; PMM: Parotid Malignant Myoepithelioma; PBM: Parotid Benign Myoepithelioma; FNA: Fine Needle Aspiration; RT: Radiation Therapy; CT: Computerized Tomography; MRI: Magnetic Resonance Imaging.

ferred for adjuvant radiotherapy after a partial parotidectomy and lymph node dissection. Although, according to the pathological report, we had negative surgical margins and no lymph node infiltration, we decided to proceed with adjuvant radiation therapy for the following reasons:

- The surgeon had expressed concerns about tumour spillage during the removal.
- There were focal regions of tumour cells infiltrating the capsule.
- There are no specific guidelines for the post-surgical treatment of a malignant myoepithelioma, but MM of the salivary glands has a relatively high tendency to recur locally [18]. There are, indeed, some reports in the literature on MMs of the head and neck region that recurred locally after successful surgical excision [10,25].
- As in this case, tumours that display marked histological aggressiveness [high mitotic rate, necrosis] may behave adversely.
- It is known that MMs arising de novo in a normal salivary gland, as in this case, tend to be more aggressive [54].

4. Conclusion

Malignant myoepitheliomas are rare tumours with no

specific guidelines for their treatment. Based on a literature review of reports of local recurrence of head and neck MMs after successful surgical excision, we believe that radiotherapy to MM of the parotid should be considered as a treatment of choice in conjunction with chemotherapy. Reported acute toxicity is minimal, while regular follow-up of the patient is necessary to report long-term toxicity of radiotherapy as well as local control of the disease. Metastases of a myoepithelioma are a rare event. As a result there is little material available as guidance and each instance should be assessed on a case-by-case basis.

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