



Mucoepidermoid Carcinoma of the Minor Salivary Glands – A Case Report and Review

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Abstract

A 27 year old woman presented to her dentist with a complaint of pain and swelling in the right retromolar area. She was referred to a local periodontist. Her symptoms persisted after extraction of tooth #32, which was believed to be the source, and subsequently failed to subside after repeated debridements and multiple courses of antibiotic therapy. The patient was referred to an oral and maxillofacial surgeon who performed a biopsy with a resultant diagnosis of an intermediate grade mucoepidermoid carcinoma. Definitive treatment consisted of a composite resection of the mandible with selective neck dissection and adjuvant radiation therapy. This case underscores the importance of recognition of the failure of a disease process to respond to standard treatment modalities and the necessity for timely referral or alteration of therapeutic course.

Case Presentation

A 27 year old African American female patient presented to her dentist in February, 2014 with a complaint of pain in the area of the right retromolar pad, with radiation to the right ear. She was referred for evaluation by a periodontist who noted swelling in the area and identified an impacted tooth #32 as the source. Teeth numbers 1,16,17, and 32 were extracted in April, 2014. Postoperatively, the pain and swelling did not subside. She returned to her periodontist for multiple local debridements were prescribed several courses of antibiotic therapy with no resolution. One year later, in June, 2015, the patient was referred to an oral and maxillofacial surgeon for evaluation. A biopsy was performed and histopathology revealed an intermediate grade mucoepidermoid carcinoma. The patient was referred to the oral and maxillofacial surgery service of Nova Southeastern University/Broward Health Medical Center for definitive treatment. Upon evaluation, the patient was found to have an erythematous and indurated swelling of the right retromolar pad which measured 1 cm x 2 cm in maximal dimension. The lesion extended superiorly along the pterygomandibular raphe to the soft palate, inferiorly along the medial aspect of the mandible, and posteriorly to the tonsil. There was no apparent involvement of the floor of mouth, tongue base, or cervical lymphatics. Computed tomography (CT) scans of the face and neck were ordered, as well as a whole body Positron Emission Tomography (PET) scan. CT of the face and neck revealed a 2.1 cm x 1.5 cm fairly well circumscribed enhancing mass in the right medial masticator space. There was no evidence of bony invasion. PET/CT identified a 1.5 cm hypermetabolic mass with a maximal SUV of 4.9. There were lymph nodes in the right jugulodigastric region which demonstrated asymmetric metabolic activity. There was no evidence of distant metastasis. After review by a multidisciplinary head and neck tumor board, the decision was made with the patient to proceed with composite resection of the right mandible and a right sided selective neck dissection. She underwent surgery on July 21, 2015. Intraoperative marginal frozen sections were free of malignancy. She was discharged on postoperative day ten after an uneventful hospital course. Final pathology was returned as a pT4N0MX intermediate grade mucoepidermoid carcinoma measuring 2.7 cm in greatest dimension with questionable superficial invasion of the bony mandible. Surgical margins were close. The cervical lymphatic contents recovered comprised thirty-eight lymph nodes, all of which were negative for malignancy. There was extensive discussion within the pathology team as to whether there was true bony invasion or whether it was due to the technique of specimen preparation. After intense deliberation, the decision was made to refer the patient to medical and radiation oncology for evaluation for adjuvant therapies. Recommendation was made for the patient to undergo radiation therapy to decrease likelihood of recurrence. It was determined that there was no role for adjuvant chemotherapy. Between November 9, 2015 and December 31, 2015 she received fractionated radiation to a total dose of 63Gy. She continues to be followed closely by our service, and at this time shows no evidence of persistent or recurrent disease.

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Discussion

Salivary glands are generally divided into the paired major glands, and the diffuse minor glands. The paired parotid, submandibular, and sublingual glands comprise the major glands, with minor salivary glands distributed throughout nearly the entirety of the oral mucosa. Notable areas of exception are the anterior hard palate and the attached gingival [7]. Benign and malignant tumors of the salivary glands occur at an annual rate of approximately [1-6]. 5 cases per 100,000 individual are worldwide [8]. They account for approximately 5% of all newly diagnosed malignancies of the head and neck [11], and comprise less than 1% of all malignancies [6]. The frequency of tumors affecting the paired major glands demonstrates an inverse relationship with the frequency of malignancy. Tumors are most commonly diagnosed in the parotid, followed by the submandibular and sublingual glands. Conversely, tumors are most frequently malignant in the sublingual gland, followed by the submandibular and parotid glands, respectively. Several large studies have determined that the minor salivary glands represent the second most common site of salivary tumor development, accounting for 9-23% of salivary tumors, and with a nearly 50% rate of malignancy [8]. The palate is the most common site of minor salivary gland neoplasia, with the retromolar tissues, floor of mouth, and tongue being least common. First described by Stewart in 1945 [11], the mucoepidermoid carcinoma is the most common malignancy of the parotid and minor salivary glands. It manifests most often during the second to seventh decades of life [8] and exhibits a slight female predilection. The most frequent presentation is a complaint of asymptomatic swelling; however, special consideration must be given to the clinical appearance of intraoral tumors associated with the minor glands. These may manifest as a blue or red hued fluctuance, resulting in clinical misdiagnosis of the tumor as a mucocele. Notably, it represents the most common salivary neoplasm affecting the lower lip, floor of mouth, tongue, and retromolar pad. The overall rate of occurrence as of 2009 was 0.7 cases per 1 million person-years [2]. Mucoepidermoid carcinoma is histologically characterized as low grade, intermediate grade, or high grade based upon assessment of cyst formation, degree of cytologic atypia, and relative numbers of mucous, epidermoid, and intermediate cells. Low grade tumors are most common, with inconsistent frequency of intermediate and high grade tumors across studies [9-11]. The palate is the most common anatomic site affected, and lesions in this site are found to be low grade in approximately 60% of cases [10]. In their review 546 cases of minor salivary gland tumors, 58.4% of mucoepidermoid carcinomas to be low grade, 38.4% to be intermediate grade, and 3.2% to be high grade. In the retromolar fossa, there was a slight predilection for low grade lesions over intermediate grade (54.5% vs. 45.5%), with no cases of high grade histology [10]. Tumor size is comparable between lesions with low and intermediate grade histology, with larger tumors associated with high grade histology [2]. Incidence of regional lymph node metastasis correlates well with histological grade, with nodal involvement in 24%, 30%, and 56% of low, intermediate, and high grade tumors, respectively. In a study exclusively assessing neck dissections performed in clinically NO patients, occult metastasis was found in 10% of patients with intermediate grade tumors and 35% of those with high grade tumors [5]. Surgical ablation is the mainstay of treatment for mucoepidermoid carcinoma, with neck dissection performed when indicated by clinically evident nodes, tumor location, or tumor size [3,8]. Indications for postoperative radiation therapy in the treatment of salivary malignancies include T3 and T4 tumors,

close margins, incomplete resection, bone involvement, perineural invasion, and recurrence. Importantly, in the case of perineural invasion of a major nerve, the cranial nerve pathway to the base of skull must be included in the field of treatment [13]. Hosokawa et al. [4] report comparable survival rates in mucoepidermoid carcinoma patients with negative margins and in those with positive margins treated with adjuvant radiation. Differential success based upon dosage was also reported, with a relapse rate of 13% in patients receiving 50Gy or less, and zero recurrences in patients receiving more than 50Gy. Generally offered only to patients with unrespectable disease, primary radiation therapy may offer a 5 year local control rate of 50% with a dosage of 66-70Gy [12]. Survival rates are highly correlated with both TNM staging and histopathology grading; however, distant metastasis has been reported even with low grade lesions [4,9]. Disease specific survival rates are variable in the literature. McHugh et al. [5] reported 5 year disease free survival of 88.3% for low grade, 90.7% for intermediate grade, and 43.1% for high grade lesions in their review of 125 patients. In their population level study of 2400 patients with mucoepidermoid carcinoma, Chen et al. [2] reported disease specific survival rates of 98.8%, 97.4%, and 67% for low, intermediate, and high grade lesions, respectively. Notably, deaths from mucoepidermoid carcinoma are more common from distant metastasis than from local or regional recurrence.

Conclusion

Salivary malignancies are relatively rare on the spectrum of intraoral disease encountered in the typical private dental practice. However, as with other forms of pathology, these aggressive lesions often masquerade as more common and benign disease processes. It is imperative for the practitioner to keep in mind that everyday diseases most often respond to everyday treatment modalities. Failure of a lesion to respond appropriately to treatment should be a “red-flag” that a more sinister process may be developing, and should warrant timely biopsy or referral for appropriate management.

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