

Unusual Case of Mucoepidermoid Carcinoma of Sublingual Salivary Gland in an Adolescent Boy

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Abstract

Mucoepidermoid carcinoma is a very commonly seen malignant neoplasm of major and minor salivary glands. It most commonly arises within the parotid gland, and one-third arises in the minor salivary glands. Sublingual salivary gland malignancies are very rare and constitute 0.5% and 1% of all tumors. The atypical location and appearance of low grade mucoepidermoid carcinomas can mislead clinicians. In this article, we describe a case of a low-grade MEC in a 13-year-old boy arising from the sublingual salivary gland which was mistaken as a ranula preoperatively.

Keywords: Malignant Salivary Gland Tumor; Ranula; Mucoepidermoid Carcinoma

Introduction

Mucoepidermoid carcinoma is a very commonly arising malignant neoplasm of the major and minor salivary glands [1]. It is the second most common malignancy [4]. Volkman described these tumors in 1895 which was further elaborated by Stewart, *et al.* in 1945. It was named as Mucoepidermoid carcinoma in 1953 by Foot and Frazell. It most commonly arises within the parotid gland and about one-third arises in the minor salivary glands. The atypical location and appearance of low grade mucoepidermoid carcinomas can mislead clinicians. In total of the head and neck malignancies, salivary gland tumors account for less than 5% [2]. Sublingual salivary gland neoplasms are extremely rare and comprise only 0.5% to 1% of all tumors. Metastasis at the time of diagnosis is seen in less than 5% cases of mucoepidermoid carcinoma [3]. Though rare, approximately 80 - 90% of sublingual gland tumors are malignant of which the most common is adenoid cystic carcinoma. Mucoepidermoid carcinoma has both epidermal cells

and mucous cells. MEC constitutes for less than 10% of all salivary gland tumors of which 2/3 arise within the parotid gland and 1/3 arise within the minor salivary glands. It is seen in any age group but is predominant between the third and sixth decade of life. Women are observed to have more predominance when compared to men (3:2). In this article, we describe a case of a low-grade MEC in a 13-year-old boy arising from the sublingual salivary gland which was mistaken as a ranula preoperatively. Similar cases were also reported by Pavani, *et al.* [13] and Aruna Kumari, *et al.* [14].

Case Report

A 13-year-old boy from Kolkata was brought to our OPD by his mother with complaints of swelling in the floor of mouth for the past 2 months. There was no history of pain, discharge or difficulty in swallowing. They already had a CT scan neck which did not show any mass lesion. On examination the swelling was soft too hard on palpation. Hence a clinical diagnosis of ranula was made

and the patient was taken up for surgery. On table, a deeper mass was seen. This mass was well circumscribed, solid and pinkish in appearance. A frozen section was sent to rule out ectopic thyroid. Frozen section was suggestive of malignancy. Hence a complete excision of the mass was done leaving free margins. The post-operative histopathology was reported as mucoepidermoid carcinoma of the sublingual salivary gland. The slide showed nests and sheets of epidermoid cells. There were also mucous cells seen embedded in epidermoid cell nests. A PET scan was done which did not show any active lesions anywhere in the body. The child is on a follow up for the last 6 months and has not shown any signs of recurrence.

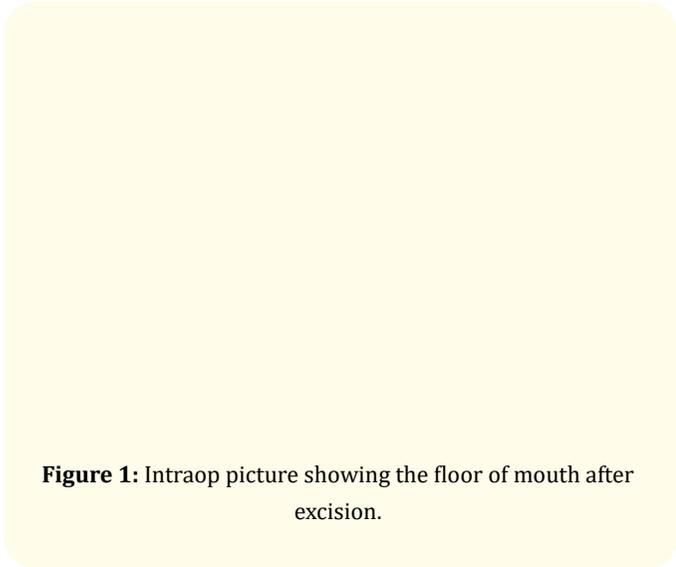


Figure 1: Intraop picture showing the floor of mouth after excision.

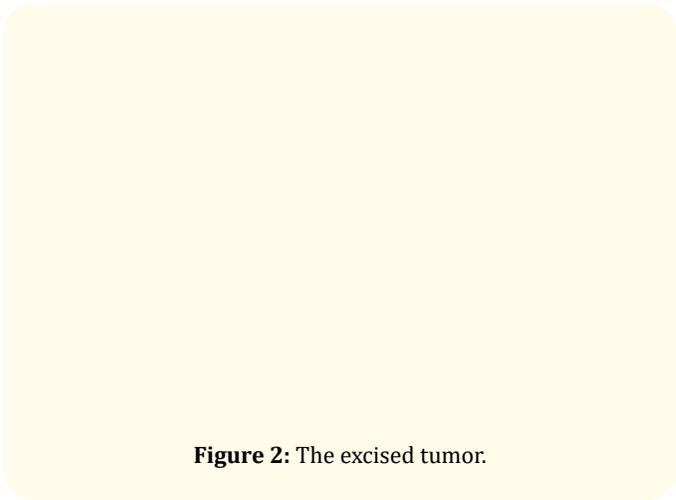


Figure 2: The excised tumor.

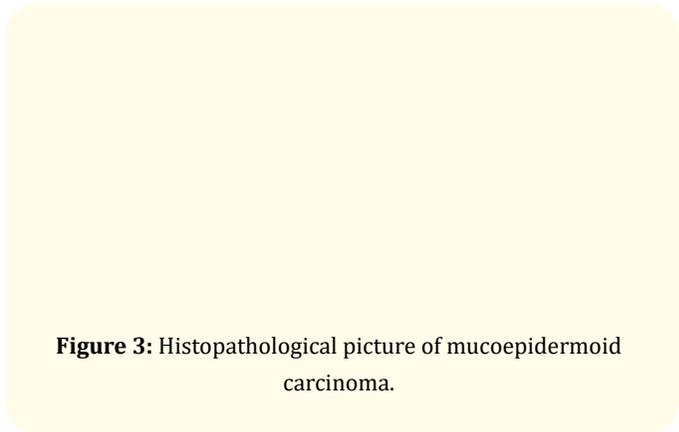


Figure 3: Histopathological picture of mucoepidermoid carcinoma.

Discussion

MEC arises from pluripotent reserve cells of the excretory ducts of salivary gland which have the potential to differentiate into squamous, columnar and mucous cells. There are no etiologic factors associated with MEC but a few cases reported of exposure to ionizing radiations [2]. The clinical presentation of MEC of the minor salivary gland is not very accurate and presents as a bluish, fluctuant, smooth surfaced mass that is often clinically mistaken as mucocele or ranula. In most of the cases MEC is seen as an asymptomatic swelling in the floor of the mouth with mild discomfort. Sometimes the tumor may even be discovered by a dentist on routine examination [6]. A similar case of a asymptomatic mucoepidermoid carcinoma was reported by Sankar, *et al.* [5]. Macroscopically MEC's are seen as small, partially encapsulated lesions. Microscopically the tumors are seen to have more mucous-producing cells than epidermoid and intermediate cells. The hallmark features of these tumors are prominent cystic structures lined by mature mucous, intermediate, or epidermoid cells, solid areas are not evident and prominent fibrous stroma is often present. It grows in a well-circumscribed manner, with no infiltrative islands at the tumor border [7]. Ranula's have more muciphages and neutrophils. MEC's have a diverse biologic behaviour and clinical presentation, which helps us stage and grade the tumor. High grade MEC is a highly aggressive tumor, whereas low-grade shows more benign nature. Metastasis can be lymphatic (cervical lymph node) or can have an haematogenous spread also. The most common site for the metastasis is lungs and the least involved is bones. Other places of metastasis are liver, brain, skin, ovary, peritoneum, etc. [8]. MEC most frequently arise in the parotid gland, their distribution being

approximately 50% in parotid gland, 40% in submandibular gland, 7% in sublingual gland and 3% in minor salivary glands. In our case floor of the mouth is involved and only a few cases have been reported involving this site [9]. The prognosis is based upon the clinical staging and histological grade. The prognosis of MEC in children is good as majority of them are well differentiated or grade I tumors. Low grade MEC cases are seen to have a better 5-year survival rate from 92 - 100% compared to high grade MEC with 0-43% survival rate [7] with an overall incidence of lymph node involvement ranging from 18 - 28% [10]. An average survival of 2.3 years and 2.6 years respectively have been observed in cases of minor salivary gland tumors with metastasis and major salivary gland tumors with metastasis [11]. Surgical resections are the main stay of treatment for malignant salivary gland tumors. The type of resection depends on the extent of the primary tumor. In case of small tumors restricted to the floor of the mouth, a wide surgical resection of involved sublingual salivary gland is done along with resection of the ipsilateral submandibular salivary gland. This is done as the ductal system is often affected with limited resection [12-14]. In the present case of low grade MEC arising from the floor of the mouth, the lesion was surgically excised along with lymphnode and clear margins were given.

Conclusion

Most of the times atypical locations and silent nature of the tumor such as in this case can mislead clinicians to make an erroneous diagnosis. This case emphasizes about the nonconfirmatory nature of plain CT and the importance of histopathological examination as the gold standard in the confirmation of diagnosis. Also, this also tells us that a routine plain CT may not always be confirmative, hence a contrast study is mandatory in suspected or doubtful cases.

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