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Case Note

Mucoepidermoid carcinoma: "A Mimicker"?

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Abstract

Neoplasms of the sublingual salivary glands are very rare and comprise only between 0.5% and 1% of all epithelial salivary tumors and approximately 1.5% of all carcinomas of the major salivary glands. Mucoepidermoid carcinomas were first described by Masson and Berger in 1924. This paper reports a case of well differentiated mucoepidermoid carcinoma in the floor of the mouth which clinically and cytologically mimicked the tumour of perineural invasion,adenoid cystic carcinoma.

Key words: Mucoepidermoid carcinoma, Salivary malignancies

Introduction

Mucoepidermoid carcinoma (MEC) is considered the most common malignant salivary gland tumor accounting for approximately one third of all salivary malignancies[1]. It consists of both epidermal and mucous cells and hence the name. Histopathologically, this tumor is classified as of either a high grade or a low grade, depending upon the ratio of epidermal cells to mucous cells. The low grade tumor has a higher ratio and is a less aggressive lesion while high grade form is considered to be more malignant tumor and has a poorer prognosis[2]. MEC is the most common salivary malignancy seen in children. Within the oral cavity, mucoepidermoid carcinoma often resembles an extravastation or retention-type mucocele and adenoid cystic carcinoma[3]. They differ from the above based on their growth pattern and mode of spread respectively. The main therapeutic method in the treatment of MEC, like in most types of salivary gland malignancies, is surgical resection. Wide surgical excision, neck dissection, and postoperative radiotherapy seem to be efficient to achieve local and regional control of the disease[4]. Here we present case of well differentiated mucoepidermoid carcinoma in the floor of the mouth which was initially thought to be adenoid cystic carcinoma.

Case Report

A 45 year old lady reported of a swelling in the floor of the mouth(Fig. 1) of 2 months duration. According to her it was sudden in onset and gradual in progression, associated with dull intermittent pain since 1 week. Patient did not report of any discharge, fever or weight loss. She was a known case of diabetes and hypertension. On examination, a single left submandibular lymphnode was palpable which was firm in consistency, mobile in all direction and non tender. Intraorally, solitary, oval swelling measuring about 5x2 cm was seen in the floor of the mouth extending to cross the midline from the first premolar on the right side to first premolar on the left side. Surface appeared smooth and normal in color. On palpation, the relevant inspectory findings were confirmed. Swelling was non tender, variable in consistency (firm to soft) with indurated borders and fixity was evident. Based on the clinical findings, a differential diagnosis of adenoid cystic carcinoma was given. Investigations included mandibular anterior topographic occlusal view, fine needle aspiration cytology, incisional biopsy (followed by histopathological analysis), and Chest x-ray to rule out metastasis.

Mandibular anterior topographic occlusal radiograph showed well defined radiolucency extending from first premolar on the right side to canine on the left side with no evidence of any calcification, suggestive of loss of labial and lingual cortical plates (Fig. 2). Chest x-ray did not reveal any abnormality. (Fig. 3)

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FNAC was done which revealed clusters and scatters of uniform cells with round to oval hyperchromatic nuclei and scanty cytoplasm. Back ground showed basophilic stromal material with hemorrhage suggestive of adenoid cystic carcinoma. For confirmatory diagnosis, histopathological examination was done and contrary to the FNAC report, it revealed acini of normal salivary gland infiltrated by tumor; tumor tissue arranged in sheaths with angulated border and eosinophilic cytoplasm with large pleomorphic nuclei ; and nerve bundles surrounded by tumor cells suggestive of high grade mucoepidermoid carcinoma. (Figures 4, 5, 6).

As a treatment modality, wide excision of the floor of the mouth along with dorsal tongue with segmental mandibulectomy and bilateral selective neck dissection was done and regularly on follow-up.

Fig. 1: Swelling in the floor of the mouth

Fig. 2: Mandibular anterior topographic occlusal radiograph showing ill defined radiolucency roughly extending from first premolar on the right side to canine on the left side

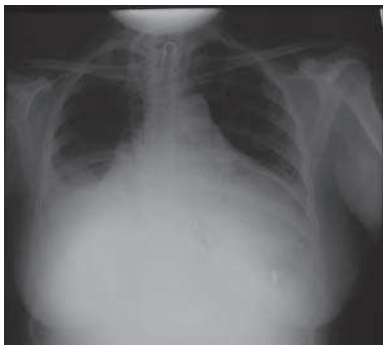

Fig. 3: Normal Chest x-ray

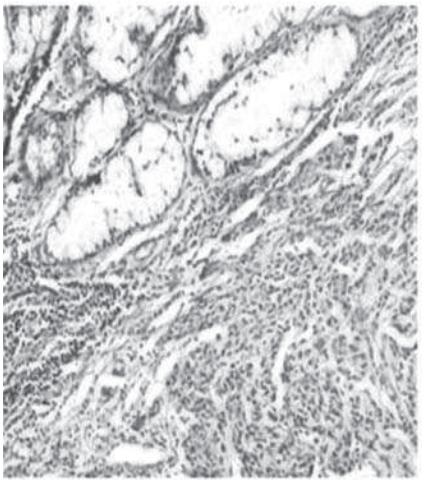

Fig. 4: Histopathological picture of mucoepidermoid carcinoma showing acini of normal salivary gland infiltrated by tumor

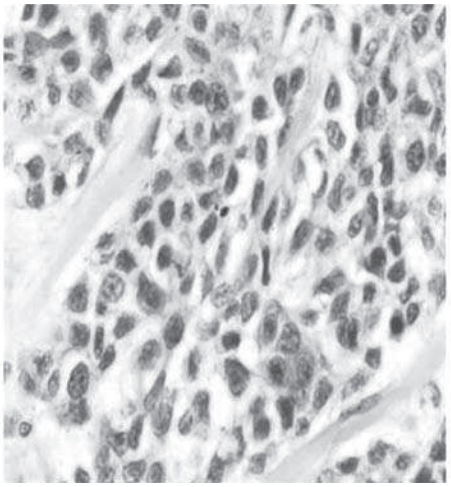

Fig. 5: Tumor tissue arranged in sheaths with angulated border and eosinophilic cytoplasm with large pleomorphic nuclei

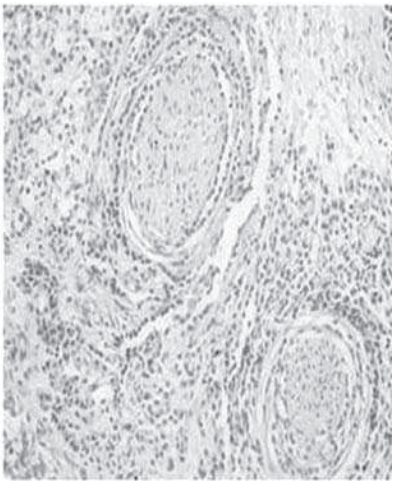

Fig. 6: Nerve bundles surrounded by tumor cells respectively (H&E × 50)

Discussion

Neoplasms of the sublingual salivary glands are very rare and comprise only between 0.5% and 1% of all epithelial salivary tumors and approximately 1.5% of all carcinomas of the major salivary glands[5,6]. Despite their rarity, approximately 80-90% of sublingual gland tumors are frankly malignant[7]. The most common malignant tumor of the sublingual gland is the adenoid cystic carcinoma[8,9]. Mucoepidermoid carcinoma (MEC) is the second most common malignancy[9,10]. Seventy percent of all salivary gland tumors are located in the parotid gland; the palate is the second most common site. Men and women are equally affected by this tumor, and the highest incidence occurs in the third to fifth decades of life[10]. Usually the presenting complaint of MEC is an asymptomatic swelling in the floor of the mouth - a mass under the tongue causing some degree of discomfort. Other symptoms include pain, trouble with dental prosthesis retention and tongue numbness. In other cases the tumor may be incidentally discovered by a dentist[11]. MEC is well known to display a widely diverse biologic behavior and a variable clinical manifestation which seems to correlate with tumor stage and grade. High-grade MEC is a highly aggressive tumor while its low-grade counterpart usually demonstrates a more benign nature[3,12]. Macroscopically, low grade MEC are usually small and partially encapsulated. High grade tumors are less likely to demonstrate a capsule because of rapid growth and local tissue invasion. Microscopically, the low grade lesions consist of regions of mucoid cells with interspersed epithelial strands. The high grade tumors consist primarily of epithelial cells, with very few mucinous cells[2]. Distant metastasis in MECs implicates an unfavorable prognosis although the biological behavior of the metastatic deposits have a slow progression[13]. When distant metastases develop in patients with minor salivary gland tumors the average survival is 2.3 years and in those with tumors of the major salivary glands is 2.6 years[1]. The lung is the most commonly involved site of metastasis[14].

Surgery remains the main modality of treatment for malignant sublingual gland tumors, although there are different types of surgical interventions according to extent of the primary tumor. For small tumors restricted to the floor of the mouth, a wide surgical resection has been suggested, including the involved sublingual and also the ipsilateral submandibular salivary gland, because the ductal system is often affected even with limited resection[15]. In tumors larger than 2.0 cm, a more aggressive approach has been recommended, using en-bloc resection. Additionally, resection of the lingual nerve and marginal mandibulectomy are necessary when there is involvement of this nerve and of the periosteum, respectively[16]. In cases with unequivocal mandibular involvement, a segmental mandibulectomy must be done. Although the risk of lymph node metastasis is low, a selective cervical lymphadenectomy is useful and it can be included in the surgical procedure without difficulties. Adjuvant postoperative radiotherapy (RT) has been indicated for patients with advanced clinical stage or high-grade tumors or when surgical margins are closed or involved by tumor[15,16]. Proper reconstruction of the floor of the mouth after surgery, especially after a marginal or a segmental mandibulectomy, is one of the major challenges for the surgeon. The current use of microvascular free-flaps has increased the function and outcome of the patients. For marginal mandibulectomy or wide surgical excision of the floor of the mouth, radial forearm or liberal arm flap are the most used. In patients where it is necessary to perform a segmental mandibulectomy, reconstruction by microvascular fibular free-flap is the most used option[15]. Local recurrence is low[16], however, with high grade lesions, recurrence of metastasis can recur in upto 60% of patients. The survival rate for patients with low-grade lesions is about 95% at 5 years; for patients with high grade lesions, this rate drops to approximately 40%[10,2].

Conclusion

A malignant tumor of salivary gland like mucoepidermoid carcinoma or adenoid cystic carcinoma, even though does not vary much in the clinical and cytological picture,can have variations in the mode of spread. Our case gives us an insight into the non confirmatory nature of FNAC and stress on the importance of biopsy as the gold standard in the confirmatory diagnosis. Recognizing an abnormality is the first and most essential step when diagnosing a disease entity. Professional responsibility and liability for recognition of abnormality and subsequent diagnosis should be emphasized.

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