EUROPEAN ENDODONTIC JOURNAL

CASE REPORT

Maxillary Mucinous Adenocarcinoma Mimicking a Lesion of Endodontic Origin: A Rare Case Report

Kavita DUBE,[1] Anjaneya DUBE,[2] Preeti JAIN,[2] Sayantan GHOSH,[2] Bonny PAUL,[1] Nupur BHATNAGAR[1]

[1]Department of Conservative Dentistry & Endodontics, Hitkarini Dental College, Jabalpur, India
[2]Department of Surgical Oncology, Saptrishi Hospital and Cancer Center, Jabalpur, India

ABSTRACT

Periapical lesions of endodontic origin are fairly common in the oral cavity in association with tooth pulp infection. Most of these lesions will resolve with adequate root canal treatment and rarely cause suspicion of more insidious disease. Most clinicians tend to skip histopathological examination in cases where the lesion is excised or curetted. We present a rare case of mucinous adenocarcinoma in association an endodontically treated maxillary discoloured central incisor in a 38 year old patient with a history of root canal treatment about 15 years ago. Root canal re-treatment and wide excision was performed. Histology showed epithelial islands suggestive of a neoplasm. Immunohistochemistry was positive for CK7 and S100. Metastasis was ruled out and no evidence of recurrence has been noted in the 12-month follow up period. It is emphasized that any tissue removed from the surgical site should be analysed microscopically.

Keywords: Mucinous adenocarcinoma, root canal treatment, surgery

HIGHLIGHTS

- Lesions associated with the endodontically treated tooth are not necessarily of inflammatory origin
- Any tissue removed from the surgical site should be analysed microscopically.
- Mucinous adenocarcinoma, although rare, may occur in the oral cavity.
- Immunohistochemistry aids in differentiating whether the tumour is of primary origin or metastatic.

Please cite this article as: Dube K, Dube A, Jain P, Ghosh S, Paul B, Bhatnagar N. Maxillary Mucinous Adenocarcinoma Mimicking a Lesion of Endodontic Origin : A Rare Case Report. Eur Endod J 2023; 8: 101-4

Address for correspondence:
Anjaneya Dube
Department of Surgical Oncology, Saptrishi Hospital and Cancer Center, Jabalpur, India
E-mail: drkdube@gmail.com

Received January 20, 2022,
Revised June 12, 2022,
Accepted October 19, 2022

Published online: January 23, 2023
DOI 10.14744/eej.2022.29200

INTRODUCTION

Tumours of the minor salivary glands are uncommon, representing 9-23% of all salivary gland tumours (1). Malignancies of the salivary glands are even more uncommon than benign tumours (2). In a study by the WHO of 92,800 biopsy samples the most common intra-oral salivary gland tumour was mucoepidermoid carcinoma (21.8%) (3). A retrospective study of 1521 biopsies found that 3.42% of cases examined did not have lesions of endodontic origin despite the clinical diagnosis of periapical inflammation, abscess, granuloma or cyst. Keratocystic odontogenic tumour (KOT) was the lesion most frequently found not to be a sequla of pulp necrosis (4). A review of 9723 biopsy reports from endodontic surgeries found that non healing apical granulomas occurred in 40.4% and cysts in 33.1% together accounting for 73% of the biopsied lesions. In addition, 8.8% were KOT and 0.3% were metastatic tumours, predominantly in the mandible (5).

CASE PRESENTATION

A 38 years old male patient reported to the Endodontic clinic with a discoloured maxillary

front tooth (Fig. 1a). He had a history of endodontic treatment about 15 years previously. On examination, the maxillary left central incisor was discoloured. The labial gingiva adjacent to the tooth appeared thickened, but had not been noticed by the patient. A hypodense lesion labial to the cervical third with an embedded bony spicule /filling material labial to the maxillary left central incisor was seen (Fig. 1b) on the cone beam computed tomography (CBCT) scan. (Carestream Dental CS 8100SC3D: 90kv: 4.00mA: 150 voxel size: FOV-5×5cm).

Considering the long-standing history and non-aggressive nature of the lesion, it was provisionally considered to be a granulomatous lesion in association with the central incisor, and the treatment plan included orthograde root canal re-treatment followed by root-end surgery to remove the lesion. The patient refused any treatment, and returned after two years with no obvious change in the clinical or radiographic picture. Written informed consent was obtained for the treatment plan as well as for submission of the removed tissue for histopathological analysis.

Root canal retreatment was performed at a private Endodontic clinic and was uneventful. The following day, after local anaesthesia, a full mucoperiosteal intra-sulcular incision was made and a trapezoidal flap raised to expose the lesion. The lesion was identified with a partial envelope of thinned out bone (Fig. 1c). The lesion was excised, aggressive curettage and peripheral ostectomy was performed (Fig. 1d, e). The lesion was sent for histopathological analysis. The flap was repositioned with sutures. Three weeks post-operative examination revealed satisfactory healing (Fig. 1f).

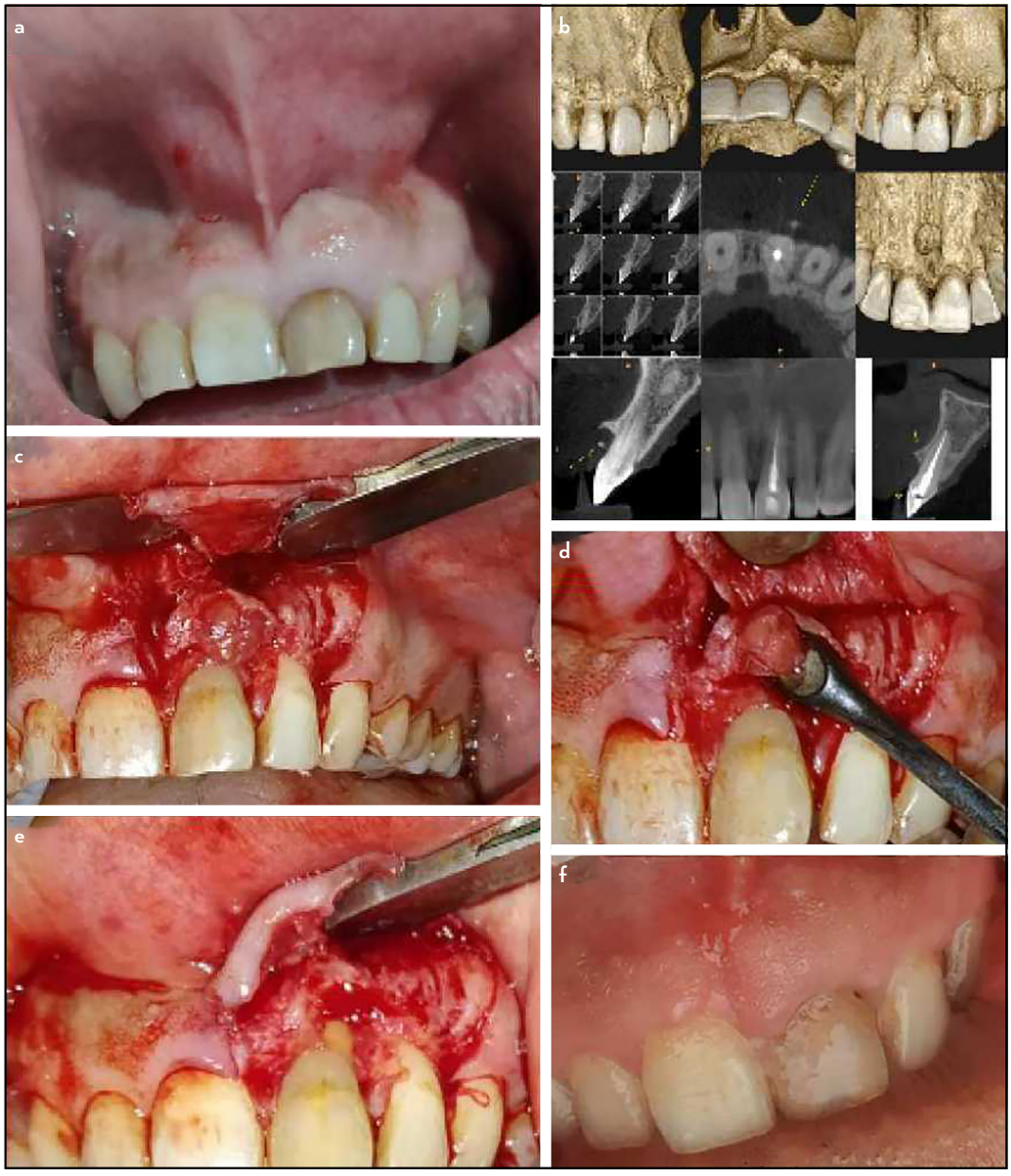

Figure 1. (a) Pre-operative photograph showing discolored upper left central Incisor. (b) Pre-operative CBCT showing hypo dense lesion labial to the cervical third with embedded bony spicule /filling material. (c) The lesion was identified with partial envelope of bone after raising a mucoperiosteal flap. (d) Excision of the lesion. (e) Surgical bed after excision, aggressive curettage and peripheral ostectomy. (f) Clinical picture at 3 week follow up

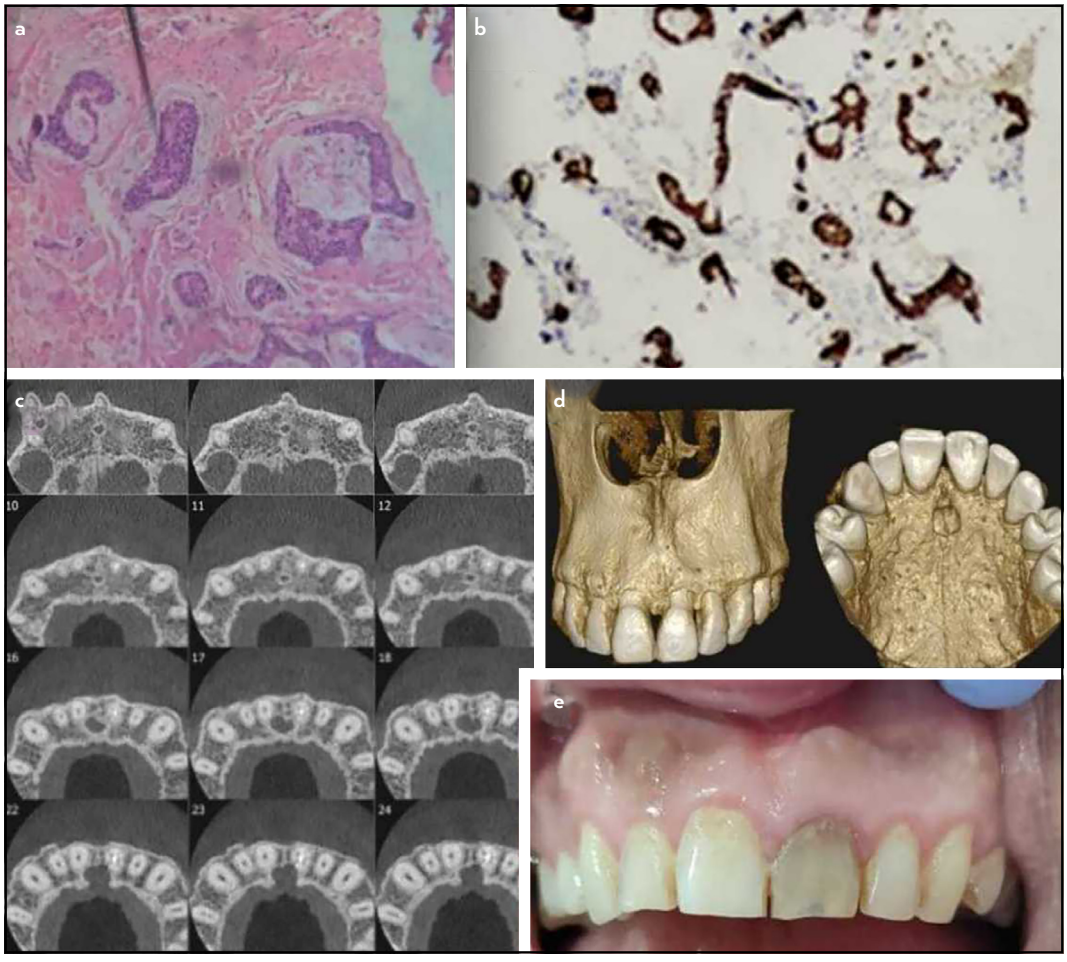

Figure 2. (a) Histopathology showed a mucosa covered tissue with underlying fibrocollagenous tissue infiltrated by moderately differentiated adenocarcinoma with extracellular mucin. (b) Immunohistochemistry showing positivity for Cytokeratin 7(CK7) and S100. (c, d) CBCT at 12-month follow up showing no recurrence. (e) Clinical picture at 12-month follow up

The biopsy showed a mucosa covered tissue with underlying fibrocollagenous stroma infiltrated by moderately differentiated adenocarcinoma with extracellular mucin. The tumour was arranged in nests, glands, trabeculae or singly scattered cells amidst extracellular mucin. Individual cells had a moderate amount of eosinophilic cytoplasm and hyperchromatic nuclei (Fig. 2a). For definitive diagnosis the blocks were sent for immunohistochemistry which showed the tumour cells to be diffusely positive for cytokeratin 7 (CK7) and S100. The findings were suggestive of mucinous adenocarcinoma favouring a primary mucosal origin (Fig. 2b).

Owing to the unusually early diagnosis with tumour size less than 0.5 cm in greatest diameter, the patient was given the options of re-excision of the tumour bed versus very close observation. Whole body metastatic work up was done at an oncology centre and no other malignancy was detected. CBCT at 12-month follow up showed no recurrence (Fig. 2c, d) and the clinical picture was also satisfactory (Fig. 2e).

DISCUSSION

The WHO defines mucinous adenocarcinoma as a malignant tumour composed of epithelial clusters within large pools of extracellular mucin (6). Mucinous adenocarcinoma of the salivary glands is a rare occurrence. It accounts for less than 0.1% of epithelial salivary gland tumours and 0.4 % of all malignancies (7). Mucinous adenocarcinoma could have a possible origin from the minor salivary glands of the labial mucosa or could possibly be metastatic carcinomas with primary origin from any other organ.

Metastatic carcinomas are rare. They contribute to about 1% of neoplasms of the oral cavity. Identification of their metastatic nature is essential as work up can then be directed towards finding the primary site, and thus potentially altering prognosis. In the present case, to exclude the presence of metastatic disease, comprehensive physical and radiological (CT scan of the head, neck, chest, abdomen and pelvis) examination were performed and no evidence of the disease was found elsewhere).

Primary mucinous adenocarcinoma cannot be differentiated from metastatic tumour based on histologic features alone. Immunotyping using CK7/CK20 can aid in determining tumour origin beside a careful clinical history and examination. The phenotype CK7+ /CK20 - is supportive of a salivary primary, whereas CK7- /CK20+ may be a clue to an intestinal origin (8). Metastasis from a remote primary is not very likely in this case, with pronounced expression of CK7. This was also supported by extensive examination and radiological tests.

The literature shows few reported cases of mucinous adenocarcinoma (MAC) manifesting clinically as gingival overgrowth. One case described peripheral MAC of the palatal gingiva with no bone involvement (9). Another case was an extensive intra- osseous minor salivary gland tumour affecting the mandibular gingiva (10).

The present case shows similarity to the reported cases in that they also resembled hyperplastic or inflammatory lesions and could have easily been misdiagnosed as granuloma or cyst. However, it is important to understand that the diagnosis of these lesions cannot be made based on the findings from any one part of the clinical or radiographic examinations. The only method to ascertain the diagnosis and differentiate it from an inflammatory lesion is a definitive histopathology.

The presentation of our case as a gingival lesion in close association with an endodontically treated tooth gave the initial impression of a lesion associated with the non-vital tooth. However, the histological findings were of a rare disease.

The tumour in the presently described case affected the labial gingiva along with the underlying bone suggestive of a central origin. Intra-osseous salivary carcinomas are very rare and occur less frequently in maxilla than mandible (11). These central tumours are thought to arise mostly from hamartomatous/ choristomatous salivary tissue entrapped within the marrow space of the jawbone during embryologic development. An alternative hypothesis regards them as arising from metaplastic odontogenic epithelium, as mucous cells can be found in various types of odontogenic cysts (10).

Due to rarity of mucinous adenocarcinoma, the information on treatment and follow up is limited. Complete surgical excision is the standard treatment. Neck dissection may be considered in high risk cases (locally advanced or neck node positive).

CONCLUSION

The findings of this case emphasize that the routine submission of resected tissue for histopathological examination is required to establish a specific diagnosis whenever tissue is removed from a surgical site. In addition to dictating further management, histopathology helps to rule out uncommon lesions. Early diagnosis of malignant tumours would enable early intervention leading to a better prognosis. Endodontists must also be aware that unsuccessful treatment, delayed response to treatment or atypical findings could be due to an unusual or uncommon disease.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Conflict of interest: The authors deny any conflict of interest.

Peer-review: Externally peer-reviewed.

Financial Disclosure: This study did not receive any financial support.

Authorship contributions: Concept – K.D.; Design – A.D., P.J.; Supervision – K.D.; Funding - A.D., S.G.; Materials - None; Data collection and/or processing – A.D., S.G., B.P.; Analysis and/or interpretation – P.J., A.D.; Literature search – N.B.; Writing – K.D.; Critical Review – B.P.

REFERENCES

1. Pires FR, Pringle GA, de Almeida OP, Chen SY. Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. Oral Oncol 2007; 43(5):463–70. [CrossRef]
2. Trattner BA, Barak Y, Tordik PA. Mucoepidermoid carcinoma mimicking a lesion of endodontic origin. J Endod 2018; 44(8):1303–7. [CrossRef]
3. Buchner A, Merrell PW, Carpenter WM. Relative frequency of intra-oral minor salivary gland tumors: a study of 380 cases from northern California and comparison to reports from other parts of the world. J Oral Pathol Med 2007; 36(4):207–14. [CrossRef]
4. Kontogiannis TG, Tosios KI, Kerezoudis NP, Krithinakis S, Christopoulos P, Sklavounou A. Periapical lesions are not always a sequelae of pulpal necrosis: a retrospective study of 1521 biopsies. Int Endod J 2015; 48(1):68–73. [CrossRef]
5. Koivisto T, Bowles WR, Rohrer M. Frequency and distribution of radiolucent jaw lesions: a retrospective analysis of 9,723 cases. J Endod 2012; 38(6):729–32. [CrossRef]
6. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. Word Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. Lyon: IARC Press; 2005. p.210.
7. Li LJ, Li Y, Wen YM, Liu H, Zhao HW. Clinical analysis of salivary gland tumor cases in West China in past 50 years. Oral Oncol 2008; 44(2):187–92.
8. Krogdahl AS, Schou C. Mucinous adenocarcinoma of the sublingual gland. J Oral Pathol Med 1997; 26(4):198–200. [CrossRef]
9. Seoane J, Varela-Centelles P, López-Niño J, Vázquez I, Abdulkader I, García-Caballero T. Gingival mucinous adenocarcinoma of a minor salivary gland. J Periodontol 2010; 81(4):626–31. [CrossRef]
10. De Benedittis M, Palmiotto A, Turco M, Petruzzi M, Cortelazzi R. Salivary mucinous adenocarcinoma of the mandible. Odontology 2017; 105(2):257–61. [CrossRef]
11. Woolgar JA, Triantafyllou A, Ferlito A, Devaney KO, Lewis JS Jr, Rinaldo A, et al. Intraosseous carcinoma of the jaws--a clinicopathologic review. Part I: Metastatic and salivary-type carcinomas. Head Neck 2013; 35(6):895–901. [CrossRef]