Clear cell variant of intraosseous mucoepidermoid carcinoma: Report of a rare entity

Sujatha Varma, PM Shameena, Sudha S, Resmi G Nair, Ipe V Varghese
Department of Oral Pathology and Microbiology, Government Dental College, Medical College PO, Calicut, India

Address for correspondence:
Dr. Sujatha Varma,
Department of Oral Pathology and Microbiology,
Government Dental College, Medical College PO,
Calicut, India.
E-mail: varma_sujatha@yahoo.com

ABSTRACT
Intraosseous mucoepidermoid carcinoma of jaw bones is a rare lesion. Abundance of clear cells in an intraosseous mucoepidermoid carcinoma may complicate its histopathologic diagnosis. It becomes extremely important to distinguish this lesion from other clear cell lesions of jaw region. Here, we report a case of clear cell variant of intraosseous mucoepidermoid carcinoma in the mandible.
Key words: Clear cells, intraosseous mucoepidermoid carcinoma, mandible, mucicarmine staining, PAS

INTRODUCTION
Mucoepidermoid carcinoma (MEC) is a common malignant salivary gland neoplasm originating in both major and minor salivary glands. It occurs mainly in the parotid gland (89.6%), followed by submandibular gland (8.4%). Intraorally it shows a strong predilection for palate. As the name implies, MEC is composed of a mixture of cell types - mucus secreting, epidermoid and intermediate cells. Clear cells are a rare finding in MECs. Clear cells may occur in focal areas or may predominate large areas of the tumor, thus complicating the diagnosis.

Though rare, MEC occurs in the jaw bones (intraosseous mucoepidermoid carcinoma) Waldron and Mustoe has classified primary intraosseous carcinomas (PIOC) in which intraosseous MEC is included as Type 4 [Table 1]. When MEC occurring in jaw bones demonstrate predominantly clear cells, the diagnosis becomes difficult. It is important to distinguish them from other clear cell lesions of the jaw region.

We report here a case of intraosseous MEC of mandible, which showed abundant clear cells.

CASE REPORT
A 50 year old male patient reported at the Department of Oral Pathology, Government Dental College, Calicut, with a painless slow growing swelling on left side of mandible at the angle – ramus region of 4 years duration. He gave a history of a similar swelling at the same location 12 years ago, which was diagnosed as dentigerous cyst associated with an impacted third molar. It was treated by enucleation and removal of the impacted tooth. The patient remained symptom free for about 7 years after the procedure, following which he developed a painless swelling, which reached the present size.

Clinical examination showed the presence of a diffuse, nontender swelling of approximately 6 × 4 centimeters over the left angle–ramus region of mandible [Figure 1]. Intraorally the swelling extended from 34 to retromolar region, obliterating the buccal vestibule [Figure 2]. The mucosa overlying the swelling was intact with normal color and smooth texture. A panoramic radiograph was taken which showed a multilocular radiolucency, which extended from 34 region toward coronoid and condyle, involving both [Figure 3]. With these features a provisional diagnosis of ameloblastoma was made. The patient underwent an intraoral incision biopsy from the lesion.

H and E stained sections of the biopsy specimen showed cystic spaces filled with eosinophilic material, surrounded by epidermoid cells and sheets of large polygonal cells with centrally placed nuclei, clear cytoplasm, and sharply defined

Table 1: Classification of primary intraosseous carcinomas (Waldron and Mustoe)

| Type   | Description                                      |
|--------|--------------------------------------------------|
| 1      | PIOC ex odontogenic cyst                         |
| 2A     | Malignant Ameloblastoma                          |
| 2B     | Ameloblastic Carcinoma                           |
| 3      | PIOC developed de novo                           |
| 4      | Central intraosseous Mucoepidermoid carcinoma    |
| a      | keratinising type                                |
| b      | non keratinising type                            |
cytoplasmic borders [Figure 4]. The intervening connective tissue stroma was scanty. The sections were stained with mucicarmine and Periodic acid Schiff’s reagent (PAS) to assess the nature of clear cells. The eosinophilic material in cyst like spaces was PAS and mucicarmine positive. Mucus-secreting cells were visualized through mucicarmine staining [Figure 5]. The clear cells retained PAS positivity after diastase digestion [Figure 6] with a focal positivity for mucicarmine [Figure 7]. Diagnosis of clear cell variant of intraosseous MEC was confirmed on this basis.

Figure 1: Clinical photograph - extra oral

Figure 2: Clinical photograph - intraoral

Figure 3: Panoramic view

Figure 4: Cyst like areas, epidermoid, and clear cells – H and E, ×100

Figure 5: Mucus cells - mucicarmine stain, ×400

Figure 6: Clear cells - PAS with diastase resistance, ×400
DISCUSSION

Intraosseous MECs though rare, tend to occur in jaw bones. Mandible is thrice more affected than maxilla. Majority of cases occur in the 4th to 5th decades of life. The clinical presentation of our case showed the classic features of intraosseous MEC.

The pathogenesis of intraosseous MEC is much debated. It may originate from:

- Entrapment of retromolar mucus glands within the mandible which undergo neoplastic transformation
- Neoplastic transformation of mucus secreting cells found in the pleuripotent epithelial lining of dentigerous cysts associated with impacted third molars.
- Developmentally induced embryonic remnants of the submaxillary gland within the mandible.
- Neoplastic transformation and invasion from the lining of maxillary sinus

Diagnostic criteria for intraosseous MEC proposed by Alexander and modified by Browand and Waldron are given in Table 2.

In addition to the typical features of MEC such as cystic spaces lined by mucus cells and epidermoid cells, an unusual finding in our case was the presence of clear cells. Clear cells may be a predominant component or rare finding in salivary gland tumors. Clear cells appear as large, polygonal cells with distinct outlines and a hydropic, water clear cytoplasm. The nuclei are small, vesicular or pyknotic, and centrally placed.

The presence of clear cytoplasm can be due to three basic factors. First, due to intracellular accumulation of nonstaining components like glycogen, lipid, or mucin. Second, due to a true scarcity of cytoplasmic organelles, and thirdly due to a fixation artifact.

The predominating presence of clear cells in otherwise definable lesions like MECs may lead to histologic misinterpretation. This necessitates the consideration of various intraosseous lesions with a clear cell component.

CONCLUSION

The clinical significance of malignant nonodontogenic tumors arising from odontogenic cysts should not be underestimated as demonstrated by the present case. This rare case of intraosseous MEC with abundant clear cells also emphasizes the need for establishment of definitive diagnostic criteria to distinguish the clear cell lesions of oral cavity and jaw bones. The use of special stains also plays an important role in diagnosis of rare lesions like intraosseous MECs showing abundant clear cells.

REFERENCES

1. Eversole LR. Mucoepidermoid carcinoma: Review of 815 reported cases. J Oral Surg 1970;28:490-4.
2. Ellis GL, Auclair PL, Gnepp DR. Mucoepidermoid carcinoma.
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In Surgical Pathology of salivary gland. 1st ed. Philadelphia: WB Saunders Company; 1991. p. 269-89.

3. Batsakis JG. Mucoepidermoid carcinoma. Tumors of the major salivary glands. In tumors of Head and Neck Clinical and Pathologic considerations. 2nd ed. Baltimore: Williams and Wilkins Company; 1979. p. 34-6

4. Yoon JH, Ahn SG, Kim SG, Kim J. Calcifications in a clear cell mucoepidermoid carcinoma of hard palate. Int J Oral Maxillofac Surg 2005;34:927-9.

5. Cawson RA, Gleeson MJ, Eveson JW. Mucoepidermoid carcinoma Pathology and Surgery of Salivary glands. 1st ed. London: Isis Medical Media; 1997.

6. Raut DL, Khedkar SA. Primary intraosseous mucoepidermoid carcinoma of the maxilla: A case report and review of literature. Dentomaxillofac Radiol 2009;38:163-8

7. Simon D, Somanathan T, Ramdas K, Pandey M. Central Mucoepidermoid carcinoma of mandible – A case report and review of literature. World J Surg Oncol 2003;1:1

8. Waldron CA, Mustoe TA. Primary intraosseous carcinoma of mandible with probable origin in an odontogenic cyst. Oral Surg Oral Med Oral Pathol 1989;67:716-24

9. Sivapathasundaram B, Bertin E. Clear cell variant of mucoepidermoid carcinoma – Report of a case and review of literature. Indian J Dent Res 2003;14:23-8

10. Rajendran R, Sivapathasundaram B. Tumors of the salivary gland Shafer’s Text Book of Oral Pathology. 6th ed. Amsterdam: Elsevier; 2009. p. 232-3

11. Neville BW, Dam M, Allen CM, Bouquot JE. Salivary gland pathology. Oral and Maxillofacial Pathology. 2nd ed. Philadelphia, PA: Saunders; 2002. p. 420

12. Alexander RW, Dupuis RH, Holton H. Central mucoepidermoid tumor (carcinoma) of the mandible. J Oral Surg 1974;32:541-7

13. Browand BC, Waldron CA. Central mucoepidermoid tumors of the jaws. Oral Surg Oral Med Oral Pathol 1975;40:631-43

14. Batsakis JG. Clear cell carcinoma: Tumors of the major salivary glands. In tumors of Head and Neck Clinical and Pathologic considerations. 2nd ed. Baltimore: Williams and Wilkins Company; 1979. p. 47-8

15. Ellis GL, Auclair PL, Gnepp DR. Mucoepidermoid carcinoma. In Surgical Pathology of salivary gland. 1st ed. Philadelphia: WB Saunders Company; 1991. p. 379-88

16. Bennington JL, Beckwith JB. Tumors of the kidney, renal pelvis and ureter, Fascicle 12. Atlas of tumor pathology. 2nd series. Washington, DC: Armed Forces Institute of Pathology; 1975. p. 148

17. Cawson RA, Binnie WH, Speight P, Barret AW, Wright JM. Clear cell odontogenic carcinoma. In Lucas Pathology of tumors of the oral tissues. 5th ed. London: Churchill Livingstone; 1998. p. 57-8

18. Slater LJ. Odontogenic Malignancies. Oral Maxillofac Surg Clin North Am 2004;16:409-24

19. Schmidt-Westhausen A, Philipsen HP, Reichart PA. Clear cell epithelial odontogenic tumor: A case report. Int J Oral Maxillofac Surg 1992;21:47-9

How to cite this article: Varma S, Shameena PM, Sudha S, Nair RG, Varghese IV. Clear cell variant of intraosseous mucoepidermoid carcinoma: Report of a rare entity. J Oral Maxillofac Pathol 2012;16:141-4.

Source of Support: Nil. Conflict of Interest: None declared.