

Primary Palatal Clear Cell Carcinoma: A Rare Case Report Highlighting Morphological Diagnostic Criteria and Surgical Management

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ABSTRACT

Background: Clear cell carcinoma (CCC) is a rare malignant epithelial neoplasm primarily arising within the minor salivary glands of the oral cavity. Characterized by an indolent clinical course, its diagnosis classically relies on identifying specific architectural and cytomorphological patterns on routine Hematoxylin and Eosin (H&E) staining, especially when immunohistochemical (IHC) profiling is unavailable.

Case Description: We report the case of a 68-year-old man presenting with a painless, slow-growing swelling on the hard palate of 3 years duration. Following clinical and radiological evaluation, left infrastructural maxillectomy was performed. Histopathological examination (HPE) on routine H&E staining revealed classic features of clear cell carcinoma arising from minor salivary gland, characterized by monomorphic cells with abundant clear cytoplasm arranged in cords and nests within a prominent hyalinized stroma. No features of a biphasic cell population or squamous/mucinous differentiation were identified.

Conclusion: While immunohistochemistry is a valuable diagnostic adjunct, classic histomorphological presentation on routine H&E remains a reliable cornerstone for diagnosing CCC in resource-limited settings or when features are highly characteristic. Wide surgical excision with negative margins offers an excellent prognosis, though long-term surveillance is necessary.

Keywords: Clear cell carcinoma, Hard palate, Minor Salivary gland tumours, Oral Oncology, Maxillectomy.

INTRODUCTION

Salivary gland tumors account for less than 3% of all head and neck neoplasms. Among these, clear cell carcinoma (CCC) - historically recognized as hyalinizing clear cell carcinoma (HCCC) - is an extraordinarily rare subtype, constituting less

than 1% of all salivary gland tumors [1]. First defined as a distinct entity by Milchgrub et al. in 1994, it predominantly affects the minor salivary glands, with the hard palate and base of the tongue being the most frequent intraoral sites [2,3].

CCC typically presents as a slow-growing, asymptomatic, mucosal-covered mass, which frequently leads to delayed diagnosis or initial misinterpretation as a benign lesion [4]. Histologically, it is defined by a proliferation of uniform cells with clear cytoplasm organized in cords, nests, or trabeculae within a hyalinized stroma [5]. When immunohistochemical data is unavailable, the primary clinical challenge lies in using a strict exclusionary morphological approach to differentiate CCC from other clear cell-dominant lesions of the oral cavity, including mucoepidermoid carcinoma, epithelial-myoepithelial carcinoma, and metastatic renal cell carcinoma [1]. This case report highlights the clinical presentation, radiological findings, and definitive H&E-based morphologic diagnosis of a primary CCC of the hard palate.

CASE REPORT

A 68-year-old male patient presented with a chief complaint of a swelling on the roof of the mouth for 3 years duration. The patient reported no history of pain, bleeding, ulceration, or paresthesia. The medical, family, and social histories were non-contributory.

Clinical Examination

Intraoral examination revealed a well-circumscribed, firm, non-tender, sessile mass measuring approximately 4x3 cm located on the Left aspect of the hard palate. The overlying mucosa was intact with central ulceration of the lesion (Fig 2). There was no clinical evidence of tooth mobility or cervical lymphadenopathy.

Diagnostic Imaging

- Computed Tomography (CT): The scan revealed a well-defined, soft-tissue mass measuring 3x2.4x2 cm localized to the hard palate. There was evidence of cortical bone erosion of the maxilla, and no deep invasion into the nasal cavity or maxillary sinus.
- Lymph Node Status: No pathologically enlarged cervical lymph nodes were identified.

Biopsy was suggestive of low-grade clear cell carcinoma.

Treatment and Surgical Intervention

Following multidisciplinary evaluation, the patient underwent a left infrastructural maxillectomy under general anesthesia. Surgical access was achieved through a modified Weber–Ferguson incision (Fig. 1). Intraoperatively, a well-defined palatal lesion with central ulceration involving the left hard palate was identified (Fig. 2).

After elevation of the skin flap, mucosal incisions were placed in the gingivobuccal sulcus and around the palatal lesion, maintaining an adequate oncological margin of 1cm (Fig. 3). Osteotomy cuts were then made through the lateral and anterior maxillary walls, hard palate, and lateral nasal wall, while preserving the orbital floor. A posterior osteotomy was completed near the maxillary tuberosity, allowing en bloc resection of the tumor-bearing segment (Fig. 4).

The surgical defect was reconstructed using an obturator prosthesis, followed by layered wound closure (Fig. 5). The resected specimen was examined grossly from both the palatal and maxillary aspects (Figs. 6 and 7) and submitted for histopathological evaluation to confirm R0 resection.



Fig 1. Modified Weber ferguson incision used for surgical access.



Fig 2. Primary lesion with central ulceration involving the left hard palate



Fig 3. Intra operative view after skin flap was raised, demonstrating gingivobuccal sulcus incision and palatal mucosal incision around tumour with adequate margin given.



Fig 4. Intra operative view of osteotomy cut through lateral and anterior maxillary wall, hard palate, lateral nasal wall. Posterior cut near maxillary tuberosity. Orbital floor is preserved.



Fig 5. Post operative appearance following obturator placement and wound closure.



Fig 6. Gross Specimen viewed from the palatal aspect.



Fig 7. Gross specimen viewed from the maxillary aspect.

Histopathological Findings (HPE)

Gross examination of the resected specimen revealed a well-demarcated, firm mass measuring ulceroinfiltrative lesion of size 3x2.5x1.5 cm not involving the maxillary bone. Microscopic evaluation of the tissue sections stained with routine Hematoxylin and Eosin (H&E) demonstrated:

- Tissue shows neoplasm composed of nests and trabeculae composed of oval to oblong shaped cells with vesicular nuclei and clear cytoplasm. Occasional mitotic figures are seen. Areas of hyalinization are seen.
- Mitotic figures were rare, and there was no evidence of necrosis, perineural invasion, or lymphovascular invasion.
- Surgical Margins: All deep and peripheral margins were confirmed negative for malignancy.

Follow-up and Outcome

The patient's postoperative course was uneventful. At follow-up, clinical examination and repeat imaging showed complete healing of the palatal mucosa with no evidence of local recurrence or regional lymph node metastasis.

DISCUSSION

Clear cell carcinoma of the minor salivary glands is classified as a low-grade malignancy [3]. While it can occur at any age, it shows a female predominance and typically peaks in the fifth to seventh decades

of life [5]. Its manifestation in the hard palate often mimics benign conditions like pleomorphic adenoma or reactive lesions, which frequently delays appropriate oncological intervention [4].

The consensus first-line treatment for localized CCC of the hard palate is wide local surgical resection with negative margins [4]. Because CCC is generally a low-grade neoplasm with an indolent course, routine elective neck dissection is not warranted in the absence of clinical or radiological nodal involvement.

However, our decision for an infrastructural maxillectomy was predicated on the need to achieve complete oncological clearance in a confined anatomical space.

- **Oncological Efficacy:** By opting for a partial maxillectomy, we effectively removed the primary tumor along with the anatomically complex palatal bone, which is often a site of potential microscopic infiltration within the palatal tissues [12].
- **Surgical Monotherapy:** The histopathological report confirmed clear margins, and given the low-grade nature of the tumor, this surgical control was deemed sufficient. This strategy prioritized the removal of the malignant burden in a single, definitive procedure, successfully sparing the patient the morbidity of postoperative radiotherapy.

The clear surgical margins achieved in this patient, combined with the low-grade histomorphology on H&E, precluded the need for adjuvant radiotherapy.

When advanced immunohistochemical profiling and molecular testing for EWSR1 gene rearrangements are unavailable, the definitive diagnosis of CCC rests entirely on identifying its classic architectural layout and systematically ruling out its histological mimics based on strict H&E morphologic criteria [1]. Recent literature highlights that "clear cell shifts" can occur across an array of oral lesions, making architectural pattern recognition critical [6]. The primary challenge on routine microscopy lies in distinguishing primary CCC from its morphologic mimics:

- Mucoepidermoid Carcinoma (Clear Cell Variant): Mucoepidermoid Carcinoma (Clear-Cell Variant): This represents a major diagnostic pitfall in palatal biopsies. Recent comparative studies emphasize that while clear cells can completely dominate the field in this variant, extensive sampling on routine H&E will eventually unmask focal intermediate cells, epidermoid nests, or faint intracellular mucin vacuoles [7,8]. The total absence of these features in our case safely excluded this differential.
- Epithelial-Myoepithelial Carcinoma (EMC): EMC typically displays a characteristic biphasic pattern consisting of an inner ductal layer of cuboidal cells and an outer mantle of clear myoepithelial cells [9]. The specimen in this case demonstrated a strictly monomorphic architecture composed entirely of sheets and cords of clear cells, lacking any dual-cell configuration.
- Clear-Cell Myoepithelial Carcinoma: Unlike the low-grade, indolent cytomorphology seen in primary CCC, myoepithelial carcinomas of the palate typically present with overt infiltrative patterns, marked cellular pleomorphism, high mitotic indices, and focal necrosis on H&E [10].

- Metastatic Renal Cell Carcinoma (RCC): Because the palate is a documented site for distant metastasis, RCC must always be considered when clear cell sheets are observed. Morphologically, RCC displays a dense, highly vascular, sinusoidal framework with prominent hemorrhage and high-grade nuclear features [9], which sharply contrasts with the dense, hyalinized fibrous stroma characteristic of primary salivary CCC.

However, long-term surveillance remains essential as late recurrences and distant metastases have been documented in the literature decades after the initial treatment [1,11].

CONCLUSION

This case report emphasizes the critical role of infrastructural maxillectomy in the surgical management of palatal malignancies. By prioritizing a wide, three-dimensional oncological resection with clear bony margins, we achieved definitive curative control of the disease in a single stage. This surgical monotherapy strategy ensured an R0 resection while avoiding the potential long-term complications associated with adjuvant radiotherapy.

Furthermore, this report underscores that while immunohistochemistry and molecular testing represent the modern standard for subtyping complex salivary gland tumors, routine H&E histopathology remains a highly dependable tool for diagnosing clear cell carcinoma when classic morphological features are present.

We concluded that surgical monotherapy with clear margins is sufficient and effective for localised CCC of hard palate and a thorough, systematic evaluation of architectural and cellular details allows for the accurate exclusion of common clear cell mimics, ensuring appropriate surgical management and long-term follow-up.

Declaration by Authors

Patient consent: Consent was obtained from the patient for publication of this case report.

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