Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Pathology

Necrotizing Sialometaplasia Following Biopsy of a Mucoepidermoid Carcinoma: A Diagnostic Pitfall

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DOI: https://doi.org/10.36347/sjmcr.2026.v14i07.010 | **Received:** 25.05.2026 | **Accepted:** 04.07.2026 | **Published:** 09.07.2026

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Abstract | **Case Report**

Background: Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor in both adults and children. Necrotizing sialometaplasia (NS) is a benign, self-limiting inflammatory lesion that is classically recognized as a histological mimic of MEC. However, its occurrence in a resection specimen following a biopsy-proven MEC represents an unusual diagnostic challenge. ***Case presentation:*** We report the case of a 15-year-old adolescent presenting with a palatal low-grade mucoepidermoid carcinoma diagnosed on incisional biopsy. Surgical excision was performed four weeks later. Histopathological examination of the entirely submitted resection specimen revealed no residual carcinoma but showed only post-biopsy changes associated with foci of necrotizing sialometaplasia. ***Discussion:*** Unlike the classical diagnostic pitfall in which necrotizing sialometaplasia mimics mucoepidermoid carcinoma, this case illustrates the opposite situation. The absence of residual tumor associated with post-biopsy necrotizing sialometaplasia may cast doubt on a previously established diagnosis of malignancy, emphasizing the importance of correlating biopsy findings with the surgical specimen. ***Conclusion:*** This report highlights an uncommon clinicopathological scenario in which post-biopsy necrotizing sialometaplasia may lead to an apparent discrepancy between the biopsy and the resection specimen. Careful histopathological review and clinicopathological correlation are essential to avoid diagnostic misinterpretation.

Keywords: Mucoepidermoid carcinoma; Necrotizing sialometaplasia; Minor salivary glands; Palate; Adolescent; Diagnostic pitfall.

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland neoplasm and the most frequent malignant salivary tumor in children and adolescents [1–4]. Histologically, it is characterized by variable proportions of mucous, intermediate, and epidermoid cells.

Necrotizing sialometaplasia (NS) is a benign inflammatory lesion of the salivary glands resulting from ischemic injury. It is of particular interest to pathologists because of its histological resemblance to malignant salivary gland tumors, especially mucoepidermoid carcinoma [5–7].

The literature has mainly focused on necrotizing sialometaplasia as a histological mimic of mucoepidermoid carcinoma. However, its occurrence as a post-biopsy change in the absence of residual tumor may create a different diagnostic challenge, emphasizing the importance of careful clinicopathological correlation between biopsy and surgical resection specimens.

CASE PRESENTATION

The study was based on histopathological specimens obtained from a 15-year-old adolescent who underwent an incisional biopsy of a palatal lesion followed by surgical excision four weeks later. Tissue samples were fixed in 10% buffered formalin, routinely processed, embedded in paraffin, and stained with hematoxylin and eosin (H&E). The entire surgical specimen was submitted for histological examination, and all slides were independently reviewed by experienced pathologists.

Histopathological Findings of the Biopsy

Microscopic examination demonstrated an infiltrative epithelial neoplasm composed predominantly of mucous cells associated with intermediate cells and fewer epidermoid cells. The tumor displayed predominantly microcystic and glandular architectural patterns. Cytological atypia was minimal, and mitotic activity was scarce. These features were consistent with a low-grade mucoepidermoid carcinoma (Figures 1 and 2).

Histopathological Findings of the Surgical Specimen

Four weeks after the diagnostic biopsy, complete surgical excision of the lesion was performed. The entire specimen was submitted for histopathological examination.

No residual carcinoma was identified despite exhaustive sampling of the surgical specimen. Histological examination revealed only post-biopsy reparative changes characterized by fibrous scar tissue and foci of necrotizing sialometaplasia, showing squamous metaplasia of salivary ducts associated with coagulative necrosis of the acinar structures. No residual malignant epithelial proliferation was identified (Figure 3).

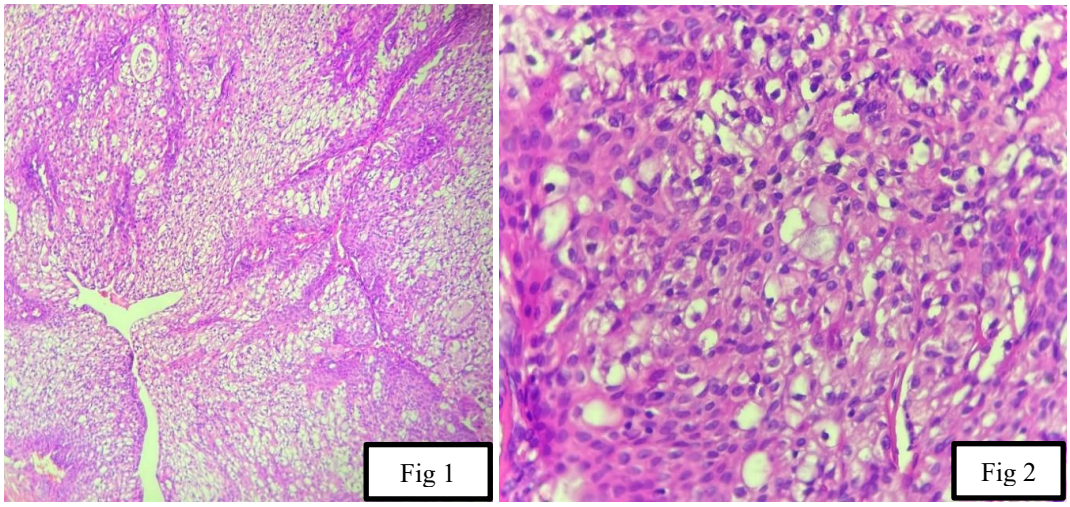

Figure 1: Low-grade mucoepidermoid carcinoma showing an infiltrative epithelial proliferation arranged in microcystic and glandular structures, predominantly composed of mucous cells admixed with intermediate cells and rare epidermoid cells (H&E, ×20)

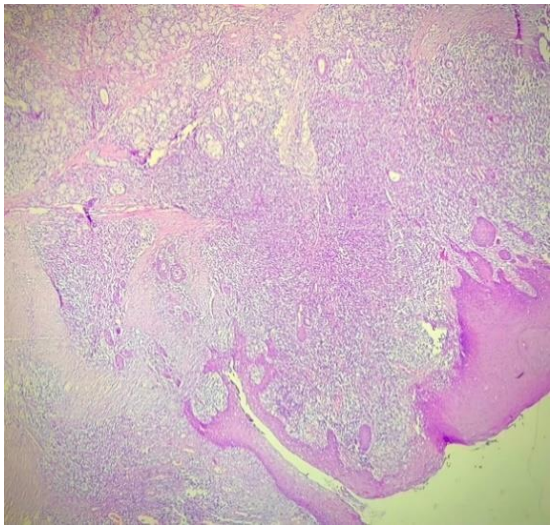

Figure 3: Panoramic view of the surgical specimen showing post-biopsy reparative changes with foci of necrotizing sialometaplasia characterized by squamous metaplasia of salivary ducts and coagulative necrosis of acinar structures, without residual carcinoma despite exhaustive histological sampling (H&E, ×20)

DISCUSSION

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland neoplasm and accounts for approximately 30% of all salivary gland carcinomas. In the pediatric population, although salivary gland malignancies are uncommon, MEC represents the predominant histological subtype. Minor

salivary glands, particularly those of the palate, are among the most frequently involved intraoral sites, and most pediatric cases are low-grade tumors associated with an excellent prognosis following complete surgical excision [1–5].

Histologically, low-grade MEC is characterized by a predominance of mucous cells arranged in cystic or microcystic structures, admixed with intermediate and scattered epidermoid cells. The diagnosis is generally straightforward when all three cellular components are identified. In our case, the biopsy showed the characteristic morphological features of a low-grade MEC, and the diagnosis was confirmed after histopathological review.

Necrotizing sialometaplasia (NS) is a rare, benign, self-limiting inflammatory condition first described by Abrams *et al.,* in 1973 [8]. It is believed to result from ischemic injury to the salivary gland lobules, leading to coagulative necrosis of the acini followed by reactive squamous metaplasia of the ductal epithelium. Several predisposing factors have been implicated, including local trauma, surgical procedures, local anesthesia, smoking, recurrent vomiting, and incisional biopsy [8–11]. Histologically, preservation of the lobular architecture despite extensive squamous metaplasia remains the key feature distinguishing NS from malignant epithelial neoplasms.

The diagnostic significance of NS lies in its well-recognized ability to mimic malignant salivary gland tumors, particularly squamous cell carcinoma and mucoepidermoid carcinoma. Because reactive squamous metaplasia may be extensive, limited biopsy specimens may lead to an erroneous diagnosis of malignancy. Conversely, recognition of preserved lobular architecture, absence of significant cytological atypia, and the accompanying ischemic changes usually allow the correct diagnosis [8–11].

The present case illustrates a distinctly different diagnostic scenario. Rather than representing a histological mimic of MEC, NS was identified exclusively in the resection specimen obtained four weeks after a biopsy-proven low-grade MEC. Despite exhaustive sampling of the surgical specimen, no residual carcinoma was identified. To our knowledge, reports describing this particular sequence of events are exceedingly rare.

Although the complete absence of residual tumor cannot be explained with certainty, complete removal of the lesion during the initial biopsy appears to be the most plausible explanation. This hypothesis is supported by the small size of most palatal minor salivary gland tumors, the low-grade nature of the lesion, and the relatively long interval between biopsy and definitive surgery, allowing the development of reparative and ischemic changes within the surgical bed. Nevertheless, this interpretation should be considered cautiously, as published evidence regarding vanishing biopsy-proven MEC is currently lacking.

The originality of this case lies in what may be considered a reverse diagnostic pitfall. Traditionally, NS raises concern because it may lead to an overdiagnosis of MEC. In contrast, our observation demonstrates the opposite situation: a genuine low-grade MEC was diagnosed on biopsy, whereas the subsequent surgical specimen showed only post-biopsy NS without residual tumor. In such circumstances, the absence of carcinoma may retrospectively cast doubt on the validity of the initial diagnosis, particularly if the biopsy slides are not carefully reviewed.

This case therefore emphasizes the importance of systematic clinicopathological correlation and direct comparison between biopsy and resection specimens. Whenever an apparent discrepancy exists, review of the original biopsy, confirmation of complete histological sampling of the surgical specimen, and integration of the clinical and surgical findings are essential before questioning a previously established diagnosis of malignancy.

Acknowledgements: None.

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