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A CASE REPORT

SQUAMOUS CELL CARCINOMA ARISING FROM A MAXILLARY RESIDUAL CYST: INTERDISCIPLINARY SURGICAL AND PROSTHODONTIC MANAGEMENT—A CASE REPORT ARISING FROM A RESIDUAL MAXILLARY CYST

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Abstract

This case report describes an 89-year-old woman diagnosed with well-differentiated squamous cell carcinoma involving a longstanding cystic lesion of the left maxilla. The clinical presentation, radiographic findings, histopathological features, surgical treatment, and prosthodontic rehabilitation are described. The patient underwent partial maxillectomy, followed by placement of a surgical obturator and subsequent interim obturator rehabilitation. Histopathological examination confirmed invasive, keratinizing squamous cell carcinoma; the report notes focal involvement of the deep surgical margin. The possibility that the carcinoma arose in a residual cyst was considered, although malignant transformation of a longstanding odontogenic keratocyst or orthokeratinizing odontogenic cyst could not be definitively excluded. This case underscores the importance of investigating persistent or atypical lesions in edentulous patients, obtaining a timely biopsy, correlating clinical and radiographic findings with histopathology, and coordinating surgical and prosthodontic care. The diagnostic uncertainty regarding the precise cystic origin and the reported margin involvement should be clearly considered when interpreting the case.

Keywords: Squamous cell carcinoma; residual cyst; odontogenic cyst; maxillary malignancy; partial maxillectomy; obturator prosthesis; prosthodontic rehabilitation.

INTRODUCTION

Squamous cell carcinoma (SCC) arising in association with an odontogenic cyst is an uncommon but clinically important event. Odontogenic cysts are frequent lesions of the jaws and are usually benign; nevertheless, their clinical and radiographic appearance may overlap with that of more consequential disease. The World Health Organization (WHO) classification places odontogenic cysts within a defined diagnostic framework and emphasizes clinicopathological correlation when distinguishing individual cyst types and related odontogenic lesions [1]. This framework is particularly relevant when a lesion presents atypically, recurs, enlarges, or is accompanied by pain, ulceration, or destructive change.

A residual cyst is generally regarded as an inflammatory radicular cyst that remains in the jaw after extraction of the associated tooth. It may be discovered incidentally in an edentulous region, and its presence does not by itself imply malignant disease. A clinicopathological study of residual cysts has described their presentation and diagnostic features, supporting the importance of careful assessment rather than relying on the clinical label alone[4]. In an edentulous patient, a swelling or radiolucency at a former tooth-bearing site can be difficult to interpret because the original dental history may be incomplete and the lesion may lack the familiar relationship to a currently present tooth. A persistent or atypical lesion therefore warrants assessment in the context of the patient's history, examination, and imaging, with tissue diagnosis when clinically indicated.

Primary intraosseous squamous cell carcinoma (PIOSCC), also referred to in some classification contexts as primary intraosseous carcinoma, not otherwise specified, is a rare malignancy arising within the jawbones. It is generally considered only after excluding

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a primary mucosal SCC extending into bone and metastatic carcinoma involving the jaws. A recent review has emphasized that the histogenesis and classification of primary intraosseous carcinoma remain challenging, in part because these tumors are rare and may resemble odontogenic cysts or other jaw lesions clinically and radiographically [2]. A 2024 report reviewing cases with cyst-lining origin likewise highlights the role of careful histopathological evaluation in establishing the relationship between a carcinoma and a pre-existing cyst [3]. These distinctions matter because the designation "SCC arising from a residual cyst" implies a specific origin that should be supported by the clinical record and histological findings, rather than inferred solely from the presence of a cyst-like radiolucency.

The diagnosis of carcinoma arising in an odontogenic cyst requires integration of clinical, radiographic, operative, and microscopic evidence. Features that raise concern include persistent or progressive swelling, pain, ulceration, irregular or poorly defined radiolucency, cortical destruction, and soft-tissue extension; however, none of these findings is individually diagnostic. Conversely, a well-defined unilocular radiolucency does not exclude malignancy. Cross-sectional imaging may help define the extent of bone involvement and the relationship to adjacent structures, but imaging cannot replace histopathological examination. Where the biopsy is limited or the epithelial lining is extensively altered, interpretation may require review by an oral and maxillofacial pathologist and correlation with the resection specimen.

Histopathological diagnosis is especially important when the lesion shows keratinizing epithelium, epithelial dysplasia, invasive growth, or a mixture of cystic and solid areas. The pathologist must distinguish invasive SCC from severe epithelial dysplasia or carcinoma in situ confined to a cyst lining and consider other keratinizing odontogenic lesions. The WHO classification recognizes odontogenic keratocyst and orthokeratinized odontogenic cyst as distinct entities; their characteristic epithelial features and biological behavior differ, so the terms should not be used interchangeably [1]. If the cyst type cannot be established confidently, the report should state that limitation explicitly. Similarly, a diagnosis of carcinoma arising in a cyst should be phrased cautiously when the available specimen does not demonstrate convincing continuity between the cyst lining and invasive tumor or when a mucosal primary cannot be excluded.

Management depends on the confirmed site of origin, tumor extent, histological findings, margin status, nodal assessment, the patient's overall health, and multidisciplinary review. Surgery may be required for resection of the involved maxilla, while further treatment decisions should be based on the complete oncologic assessment rather than on the rarity of the presentation alone. The literature on primary intraosseous carcinoma also notes that staging and treatment approaches can be difficult to standardize because the condition is uncommon and published series are limited [2]. For this reason, a case report should document the basis for the diagnostic and staging decisions and explain the rationale for any decision not to provide additional therapy when adverse pathological features are reported.

Maxillary resection can produce an oral–nasal or oral–sinus communication and may affect speech, swallowing, mastication, facial appearance, and social functioning. Prosthodontic rehabilitation with a surgical, interim, and, when appropriate, definitive obturator can help restore separation between the oral and nasal cavities while the surgical site heals. The choice between obturator rehabilitation and surgical reconstruction is individualized and depends on defect size and location, anticipated function, medical status, patient preference, and available expertise. Reporting both oncologic management and the functional rehabilitation plan is therefore valuable: it allows readers to understand not only how the lesion was treated, but also how the consequences of maxillectomy were addressed.

This case report describes an elderly, edentulous patient with invasive, well-differentiated keratinizing SCC involving a cystic lesion of the left maxilla. The report presents the clinical, radiographic, and histopathological findings and outlines partial maxillectomy followed by obturator rehabilitation. Because the precise cystic origin is not unequivocal in the original diagnostic description, the case also illustrates the importance of expressing diagnostic certainty carefully and documenting the pathological margin status, staging assessment, rationale for adjuvant-treatment decisions, and available follow-up. The purpose is to emphasize careful investigation of atypical cystic lesions and coordinated oncologic and prosthodontic management, without implying that malignant transformation is a common outcome of residual cysts.

CASE PRESENTATION

An 89-year-old woman presented with swelling in the left maxillary region accompanied by moderate pain. She had been edentulous for more than five years. Clinical examination showed complete edentulism of the maxillary arch with an ulcerative swelling of the left buccal alveolar ridge in the former tooth-bearing region of teeth #10–12. The patient reported no relevant risk factors and no previous history of malignancy. Radiographic examination demonstrated a unilocular radiolucency in the left maxilla, centered in the former region of tooth #12, extending anteriorly in the former site of tooth #11 (Figure 1).

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Figure 1. Radiographic evaluation demonstrating the unilocular left maxillary radiolucency of site #11-12.

An incisional biopsy was performed. The submitted specimen was described as a whitish-gray cystic lesion containing keratinous material (Figure 2A–B) and was sent to the Oral and Maxillofacial Pathology Department at the Medical University of South Carolina for microscopic evaluation.

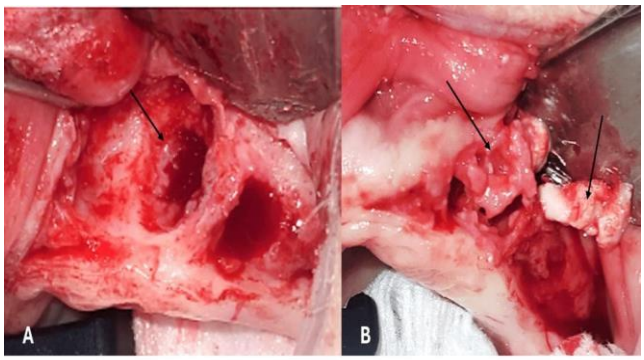


Figure 2. Intraoperative photographs of the left maxillary lesion. (A) Initial surgical reflection showing irregular cortical borders of the alveolar bone in the former site of #11. (B) Lesional tissue removed from the alveolar sites formally #11 and #12.

Histopathological examination showed multiple fragments of atypical, dysplastic keratinized epithelium, granulation tissue, fibrous connective tissue, and keratinaceous debris (Figure 3A).
The cystic lining was composed of atypical verruciform ortho- and parakeratinized epithelium, with broad, pushing epithelial fronts and bulbous rete ridges (Figure 3B).
Neoplastic cells demonstrated individual-cell keratinization, nuclear hyperchromasia, increased nuclear-to-cytoplasmic ratio, abnormal mitotic activity, and loss of cellular polarity within the middle-third of the epithelium (Figure 3C).
Keratin pearl formation was also observed. No lymphovascular or perineural invasion was identified in the examined material.

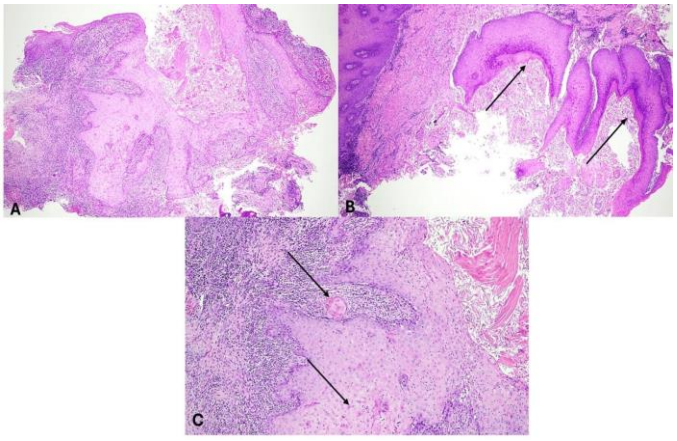


Figure 3. Photomicrographs of the biopsy specimen showing atypical, dysplastic verruciform ortho- and parakeratinized epithelium. (A) Low-power view showing an atypical cystic lining and central keratinaceous debris (H&E, ×4). (B) Higher-power view highlighting the atypical verruciform ortho- and parakeratinized cystic epithelial lining (H&E, ×10). (C) Keratin pearl formation, individual-cell keratinization, and increased nuclear-to-cytoplasmic ratios (H&E, ×10).

The final diagnosis was invasive, well-differentiated, keratinizing SCC of the left maxillary gingiva, described as arising from a residual cyst. The report states that the carcinoma extended focally to the deep surgical margin. The tumor was recorded as pT2NxMx under the AJCC/UICC TNM staging system[6]. The staging reference supplied in the original manuscript concerns salivary gland carcinoma and may not be appropriate for SCC of the maxillary gingiva; the authors should verify the applicable staging chapter and edition and confirm the basis for the reported pT category. The nodal and distant-metastasis categories should also be reported in accordance with the actual clinical work-up, rather than using “NxMx” without explanation.
The authors noted that malignant transformation of a longstanding odontogenic keratocyst or orthokeratinizing odontogenic cyst could not be entirely excluded. Accordingly, although the lesion was described as a residual cyst, the precise cyst type and the evidence supporting its origin should be stated cautiously. SCC arising in association with a residual cyst is uncommon and should be considered in the differential diagnosis of persistent or atypical cystic lesions [4].
Management was planned by an interdisciplinary team, with prosthetic rehabilitation incorporated into the surgical treatment plan.
Treatment consisted of partial maxillectomy with immediate placement of a surgical obturator. The obturator was relined intraoperatively with a soft reline material to improve its adaptation to the defect and provide a seal, with the aim of supporting comfort and speech and swallowing during early healing. Fixation screws were used to secure the prosthesis. The manuscript

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states that adjuvant radiotherapy was not recommended and that postoperative trismus was not anticipated. Given the reported focal involvement of the deep surgical margin, the basis for the adjuvant-treatment decision should be clarified, including whether the case was discussed by a multidisciplinary head-and-neck oncology team and whether further resection, radiotherapy, or close surveillance was considered.

Ten days after surgery, the surgical obturator was replaced with an interim obturator intended to support mastication, speech, swallowing, and facial appearance during healing. The size of the surgical defect and the absence of teeth presented potential challenges for prosthesis retention. The manuscript would be strengthened by providing the available follow-up duration and the patient's functional and oncologic outcomes, including whether the obturator remained stable and whether recurrence was observed.

DISCUSSION

Residual cysts are usually benign, but cystic lesions of the jaws may occasionally show unexpected or aggressive behavior. A persistent or enlarging lesion in an edentulous patient should not be assumed to be benign solely because it occurs at a previous tooth-bearing site. Clinical suspicion should increase when swelling is accompanied by pain, ulceration, an ill-defined radiolucency, or other atypical findings [4,7]. In such circumstances, biopsy and histopathological assessment are essential.

Head and neck SCC most often arises from the mucosal epithelium of the oral cavity, pharynx, larynx, or sinonasal tract, and many cases do not have a previously recognized premalignant lesion[8]. The histological development of invasive carcinoma may involve epithelial hyperplasia and dysplasia, carcinoma in situ, and subsequent invasion, although this sequence is not necessarily documented in every individual case [9]. Prognosis is influenced by tumor site and stage, patient age and general health, relevant exposures, and pathological features. Timely diagnosis and appropriate staging are therefore central to treatment planning.

Radicular cysts develop in association with inflammation at the apex of a non-vital tooth. Inflammatory mediators can stimulate proliferation of odontogenic epithelial rests within periapical inflammatory tissue, contributing to cyst formation. A residual cyst may persist after extraction of the causative tooth. However, the proposed origin of a carcinoma in a residual cyst requires adequate histopathological and clinical evidence. In the present case, the authors acknowledge that an odontogenic keratocyst or orthokeratinizing odontogenic cyst could not be excluded. This uncertainty is important and should be retained in the interpretation rather than presenting the cystic origin as definitively established.

Tobacco and alcohol exposure are major risk factors for many head and neck SCCs, although their absence does not exclude the diagnosis. SCC may present as a non-healing ulcer, an exophytic or endophytic mass, or a lesion associated with pain or bone destruction. Advanced disease may be accompanied by regional lymphadenopathy or invasion of adjacent structures. The present patient's age, pain, swelling, ulceration, and radiographic abnormality illustrate why persistent lesions in edentulous patients merit thorough assessment. The broad epidemiological statements in the original manuscript should be supported by a directly relevant, accurately cited source and should not be attributed to the National Institutes of Health unless the specific source is identified.

Cross-sectional imaging, including computed tomography or cone-beam computed tomography when appropriate, can help define lesion extent and bone involvement. Imaging does not replace biopsy for definitive diagnosis. Once malignancy is identified, treatment planning should be based on tumor site, extent, staging, margin status, nodal assessment, patient fitness, and multidisciplinary evaluation. Depending on these factors, management may include surgical resection, neck management, radiotherapy, and functional rehabilitation. In this case, the report gives a pT2NxMx classification and describes focal involvement of the deep surgical margin, but it does not provide enough information about nodal evaluation, the rationale for the final stage, or the decision not to use adjuvant radiotherapy. These details are particularly important for readers assessing the adequacy of treatment.

The original manuscript cites a transformation rate of approximately 0.12% for odontogenic cysts and suggests a higher incidence in keratinized than in non-keratinized cysts [4]. Because estimates may vary with the definition of malignant transformation, cyst type, study design, and case ascertainment, this figure should be checked against the cited source and presented with appropriate qualification. It should not be interpreted as an individual patient's risk.

Maxillectomy can create an oral–nasal or oral–sinus communication and may affect speech, swallowing, mastication, and facial appearance. An obturator is an established prosthodontic option for selected patients and can help separate the oral and nasal cavities while supporting function and appearance[7–9]. Immediate surgical obturation may assist during the initial postoperative period, followed by adjustment or replacement as the defect heals. The choice between obturator rehabilitation and surgical reconstruction, including microvascular free-flap approaches, depends on the defect, oncologic requirements, medical status, patient preferences, local expertise, and anticipated functional outcomes [8–11]. In the present case, the interim obturator was used despite the retention challenges associated with

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the size of the defect and the lack of dentition. Additional follow-up information would help demonstrate the practical outcome of this approach.

Several limitations should be acknowledged. This is a single case, so it cannot establish the frequency or causal mechanism of malignant transformation. The precise underlying cyst type remains uncertain according to the authors' own description. The reported deep-margin involvement, staging designation, and decision against adjuvant radiotherapy require clearer documentation. Finally, the manuscript does not provide a follow-up interval or detailed long-term functional and oncologic outcomes. Addressing these points would improve the report's clinical usefulness and allow readers to assess the diagnostic reasoning and management more fully.

CONCLUSION

Early recognition of oral SCC is important, and persistent or atypical cystic lesions in the jaws require careful evaluation even in edentulous patients. A non-healing ulcer, pain, swelling, or an unusual radiographic appearance should prompt appropriate investigation and, when indicated, biopsy. This case highlights the value of correlating clinical, radiographic, and histopathological findings and of coordinating surgical treatment with prosthodontic rehabilitation. At the same time, the diagnosis of carcinoma arising specifically from a residual cyst should be presented in light of the stated differential diagnosis, and the staging, margin status, adjuvant-treatment decision, and follow-up outcomes should be documented clearly. A multidisciplinary approach is essential for individualized oncologic management and functional rehabilitation.

DECLARATIONS

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Competing and Conflicting Interests

The authors declare that they have no competing or conflicting interests.

Ethical Approval

All procedures involving the patient were performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

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