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Tumor Discovery

CASE REPORT

High-grade sinonasal adenocarcinoma as an unusual presentation: A case report

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Abstract

The occurrence of malignant tumors, such as adenocarcinoma, is commonplace in the nasal region, characterized by a notably low incidence. This rarity often contributes to delayed diagnosis, rendering the condition challenging to manage. Here, we present the case of a 70-year-old man who presented with chronic left nasal obstruction, accompanied by headaches and recurrent epistaxis. The diagnostic investigation revealed an extensive and aggressive nasal adenocarcinoma, necessitating a hemi-maxillectomy for tumor excision. Subsequent reconstruction of the ipsilateral cheek was accomplished using a pectoralis major flap. Despite the initial intervention, the patient experienced a recurrence 1 year later. In sinonasal adenocarcinoma, nasal obstruction stands out as the predominant symptom, complemented by potential manifestations of swelling and facial deformation. The inherently aggressive nature of this tumor underscores the imperative for early diagnosis and meticulous management to achieve optimal therapeutic outcomes. The comprehensive approach involving surgical intervention and reconstruction reflects the complexity in treating advanced nasal adenocarcinomas.

Keywords: Adenocarcinoma; Malignancy; Nose; Paranasal sinuses

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1. Introduction

Primary sinonasal adenocarcinomas are rare tumors exhibiting a broad morphological spectrum. They are generally divided into two groups: intestinal-type sinonasal adenocarcinoma (ITAC) and non-ITAC subtypes.[1] These neoplasms are estimated to constitute merely 13% of all documented cases of sinonasal carcinoma.[2,3]

The classification of adenocarcinoma holds paramount significance as it not only dictates the tumor's behavior but also predicts the patient's prognosis. Observational data indicate that individuals afflicted with low-grade adenocarcinomas typically endure prolonged symptoms, experience reduced pain, and are less prone to deformities. Conversely, high-grade lesions entail more extensive involvement of the paranasal sinuses, signifying heightened invasiveness.[4]

The primary objective of implementing surgery is to completely remove the lesion to ensure a favorable outcome. While various external approaches can be employed, endoscopic surgery has gained prominence in recent years. This shift is attributed to its comparable clinical outcomes, lower incidence of complications, and diminished

mortality rates in contrast to alternative surgical procedures.[5,6] Radiotherapy pre-dominantly serves as a palliative or complementary measure in the post-operative phase.[5,7] Despite its positive effects, chemotherapy has waned in popularity due to its suboptimal reproducibility of therapeutic effects. Evidently, surgery remains the most advantageous treatment modality, whether utilized independently or in conjunction with radiotherapy.[5] Adjuvant radiotherapy is recommended for high-grade tumors and those classified as T3 or T4 stage.[6]

The case under consideration involves a 70-year-old patient diagnosed with high-grade adenocarcinoma of the nasal cavity, presenting initially with the left nasal obstruction. After the initial intervention, the patient became subject to a recurrence of the tumor with extension to the left cheek, highlighting the aggressive nature and metastatic potential of this subtype of adenocarcinoma.

2. Case report

A 70-year-old man, with a history of chronic smoking (30 packs/year) and occasional alcohol consumption, presented at the clinic due to persistent, unilateral left nasal obstruction, and recurrent episodes of epistaxis that had been ongoing for 6 months. The patient did not report any associated symptoms such as facial pain, anosmia, or rhinorrhea.

On anterior rhinoscopy, a notable deviation of the right nasal septum was identified. Subsequent endoscopic examination revealed a sizable, lobulated mass within the left nasal cavity, characterized by a tan, fleshy, and firm consistency, fully occupying the cavity. Despite its considerable size and where it was located, the mass did not display hypervascularization. A biopsy was conducted, confirming the presence of a glandular tumor. However, the specific origin of the tumor could not be determined through endoscopic examination due to its extensive dimensions.

Further, examination, which included the evaluation of the nasopharynx, eyes, neck, and cranial nerves, revealed normal findings. A comprehensive magnetic resonance imaging (MRI) assessment of the paranasal sinuses disclosed a massive and expansive lesion occupying the entire left nasal cavity up to the level of the choana (Figure 1). The lesion was measured at 9.0 × 4.8 × 2.1 cm in dimensions, with evident contrast entrapment, and the absence of associated lymphadenopathy. Given the increased risk of bleeding and the intent to perform an *en bloc* resection, the initial biopsy was deferred, and a decision was made to proceed directly with tumor resection.

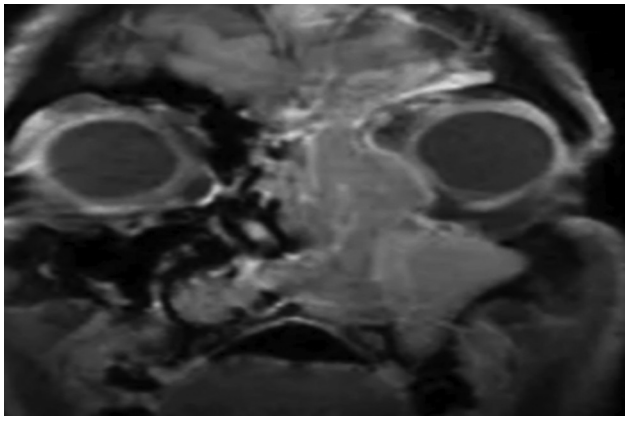


Figure 1. Pre-operative magnetic resonance imaging of the patient's nasal cavity

The patient underwent surgical intervention, which included tumor resection with hemi-maxillectomy. Preservation was undertaken for the medial orbital wall and the anterior skull base. The lesion was successfully removed and pathologically identified as a glandular tumor. Subsequent immunohistochemical analysis revealed a high-grade non-intestinal adenocarcinoma classified as pT3N0M0 (Figure 2). Postoperatively, the patient was referred to the oncology department for comprehensive oncological management due to the aggressive nature of the identified adenocarcinoma.

The patient was lost to follow-up for a year. On returning to the clinic, he suffered from a left jugal ulcerating and infiltrating mass, which was fistulized to the skin. Notably, no discernible signs of local recurrence were observed during the rhinoscopy. Figure 3 depicts the recurrent lesion, which was unfortunately neglected by the patient. The ulceration and infiltration of the jugal mass, along with the fistulization of the skin, reflect the progression of the disease due to a lack of care during the time when the patient was lost to follow-up.

A computed tomography (CT) scan revealed a well-defined left jugal tissue process measuring 69 × 62 mm, extending over 60 mm. The extension encompasses the medial aspect of the left nasal fossa, the left ramus, the left horizontal branch of the mandible, and the left zygomaticus. Posteriorly, it extended to the left masseter muscle, displaying a loss of the separation line (Figure 4). Surgical excision (exeresis) of the tumor was executed, followed by the reconstruction of the resulting defect. The reconstruction involved employing a temporalis muscle flap, complemented by a skin graft to address the loss of skin substance. The procedure also included a dissection of the homolateral lymph nodes (Figure 5).

On completion of the surgery, a definitive anatomopathological study was conducted, revealing the

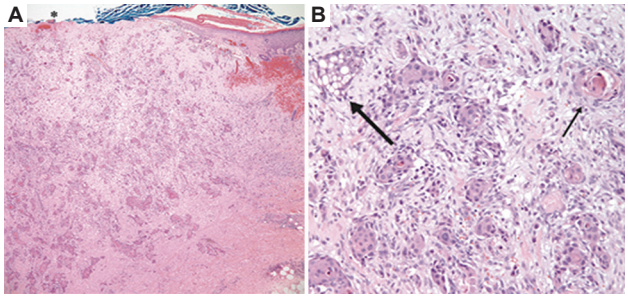


Figure 2. Characteristic histopathologic features of adenosquamous carcinoma. (A) Infiltrative pattern of neoplastic nests, dermal fibrosis/sclerosis, elastosis, and focal ulceration (denoted by asterisk) (hematoxylin-eosin staining; magnification ×40. (B) Keratinizing cysts (denoted by small arrow) and glandular elements (denoted by large arrow) (hematoxylin-eosin staining; magnification ×200.



Figure 3. Image showing tumor recurrence at the left jugal level



Figure 4. Pre-operative compared tomography scan showing well-bounded, round, and heterodense jugal tissue process measuring 69 × 62 mm

characteristic appearance of cutaneous adenosquamous carcinoma. Notably, the findings included the presence of vascular emboli and peri-nervous infiltration. Importantly, no associated lymph node metastases were identified.

Subsequent to the anatomopathological assessment, the patient was referred to the oncology department for further



Figure 5. Image showing the loss of substance after excision of the cheek mass

treatment. This referral underscores the necessity for a comprehensive oncological management plan, considering the aggressive nature of adenosquamous carcinoma and the distinctive morphological characteristics observed in the anatomopathological analysis.

3. Discussion

Glandular neoplasms account for 4 – 8% of all primary malignancies within the nasal cavity.[1,2] Reports on high-grade sinonasal adenocarcinomas of the non-intestinal type are scant.[2,9] This type of tumor is more common among male individuals and affects people across a broad age spectrum, spanning from adolescents to the elderly.[3,4] Nasal obstruction, often accompanied by swelling or facial deformities, emerges as the predominant clinical symptom. The nasal cavity and maxillary sinuses are the most frequently implicated sites, although in some cases, such as the one presented here, extension to other sinuses is possible. Unlike adenocarcinomas of the intestinal type, no discernible risk factors have been identified for high-grade non-intestinal adenocarcinomas.[5]

The prognosis for high-grade tumors is typically unfavorable, even if aggressive therapeutic interventions are implemented, associated with a meager 3-year survival rate of 20%.[6,7] Historically, the average 5-year survival rate for sinus cancer has shown a notable increase from 28% in the 1960s to 51% in the 1990s.[8] Recent studies, including an analysis by Turner and Reh, highlighted an incremental clinical improvement following sinonasal cancer treatment, with a 5-year relative survival rate estimated to have risen from approximately 49.7% in 1973 to 56.4% in 2001.[4] Choussy *et al.*'s analysis of 418 patients reports a 5-year overall survival rate of 64%.[10]



Figure 6. Image showing reconstruction with a flap of the temporalis muscle

Distinguishing between low-grade and high-grade adenocarcinomas is pivotal for treatment planning and prognostic assessments. Histologic features indicative of high-grade adenocarcinomas encompass solid growth patterns with sheets of cells, poorly defined irregular glandular patterns, hyperchromatism, moderate to prominent nuclear pleomorphism, and a heightened mitotic rate.[9,10]

Diagnostic evaluation of sinonasal masses may involve radiologic modalities such as CT and MRI. Either modality can reveal tumor involvement in various anatomical regions, including the nasopharynx, intracranial cavity, paranasal sinuses, orbits, infratemporal fossa, and pterygopalatine fossa. Determining the tumor origin on CT may pose challenges, especially in cases involving a common wall, such as the medial wall of the maxillary sinus, and the detection effort could be further compounded by obstructive sinusitis. In such instances, MRI proves to be a valuable adjunct for detection.

The primary therapeutic modality for sinonasal adenocarcinoma is complete surgical excision. In a study by Alessi *et al.* involving 13 patients, the adequacy of surgical margins emerged as the single most crucial factor in treatment success, although no specific margin size was recommended. In anatomical regions such as the superior nasal vault and skull base, achieving clear margins may necessitate multiple excisional biopsies due to the potential presence of microscopic disease, even in apparently normal-appearing mucosa. Surgical approaches for low-grade adenocarcinoma should be less radical compared to the more aggressive strategies warranted for high-grade lesions.[11,12] Adjuvant radiation therapy is recommended for high-grade lesions and recurrent low-grade lesions.[13]

Adenocarcinomas detected in their early stage can be managed with only surgical treatment, which results in an impressive 5-year survival rate of 83.4%. For patients with advanced-stage disease, the combined approach of surgery and radiotherapy, which yields a 5-year survival rate of 66.6%, is recommended. Notably, radiotherapy alone does not confer a significant benefit in terms of 5-year survival when compared with no-treatment.[14]

This case provides valuable insights, emphasizing the rarity and aggressiveness of high-grade non-intestinal sinus adenocarcinomas. Nasal congestion emerges as a prominent and non-specific symptom, often contributing to delayed diagnoses. Thus, multidisciplinary assessments play a pivotal role in effective patient management. Complete tumor resection through meticulous surgical approaches proves superior to endoscopic methods. This case report underscores the necessity to perform surgical resection to ensure optimal clinical outcomes (Figure 6).

4. Conclusion

Adenosquamous carcinoma is aptly characterized as a locally aggressive, high-risk subtype within the spectrum of cutaneous squamous cell carcinoma. Histopathological attributes of this tumor subtype, such as tumor thickness and perineural invasion, confer a heightened risk, and the incidence of locoregional recurrence is notably common. Distinguishing adenosquamous carcinoma from other sinonasal tumors is imperative due to its markedly distinct prognosis. Achieving a definitive diagnosis necessitates a comprehensive anamnesis, encompassing a detailed patient history and clinical evaluation. The diagnostic process is further refined through radical endoscopic resection, aimed at achieving a total excision of the lesion. This surgical approach is crucial for both diagnostic accuracy and therapeutic efficacy. In addition to the surgical intervention, satisfactory immunohistochemistry results play a pivotal role in confirming the diagnosis of adenosquamous carcinoma. Immunohistochemical analyses provide valuable insights into the specific molecular markers and protein expressions associated with this subtype, aiding in its conclusive identification. In summary, recognizing adenosquamous carcinoma as a distinct and aggressive subtype underscores the importance of implementing a thorough diagnostic investigation. A comprehensive patient history, radical endoscopic resection for total excision, and reliable immunohistochemistry results collectively contribute to achieving a definitive diagnosis and informing the subsequent course of therapeutic interventions.

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Conflict of interest

The authors declare that they have no competing interests.

Author contributions

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Writing – original draft: Sara Moujrid, Fadoua El Mourabit
Writing - review & editing: All authors

Ethics approval and consent to participate

Verbal consent had been obtained from the patient before participation.

Consent for publication

Verbal consent had been obtained from the patient for publishing this case report while maintaining his anonymity.

Availability of data

Not applicable.

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