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Carcinosarcoma Arising from Inverted Papilloma in a Patient with History of Radiotherapy for Sinonasal Squamous Cell Carcinoma

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 72-year-old
Final Diagnosis: Sarcomatoid carcinoma arising from inverted papilloma
Symptoms: Nasal mass • nasal obstruction
Medication: —
Clinical Procedure: Excsion
Specialty: Pathology

Objective: Challenging differential diagnosis

Background: Carcinosarcoma of the sinonasal tract is an extremely rare malignant neoplasm; it is often designated as carcinoma with spindle cell or sarcomatoid features. We report a case of carcinosarcoma arising in a pre-existing inverted Schneiderian papilloma in the left maxillary antrum and nasal cavity of a 72-year old male patient.

Case Report: The patient had a significant history of radiotherapy for squamous cell carcinoma in the sinonasal area, 3 decades ago. The patient presented with chief complaints of left nasal blockage, nasal discharge, anosmia, and occasional epistaxis. Computed tomography scan displayed a lobular soft tissue mass resulting in narrowing of the nasopharyngeal airway with massive destruction of palatal tissue. The lesion was resected via endoscopic surgery. Macroscopically, a white fleshy appearance with necrosis was noted in the submitted specimen. Microscopically, the tumor was composed of pleomorphic epithelial and spindle cells with numerous mitoses and remarkable tissue necrosis. Residual inverted papilloma (IP) with high-grade dysplasia, and minimal foci of moderately differentiated squamous cell carcinoma (SCC) component was present at the tumor margin. A distinct zone of transition of SCC to spindle cell carcinoma (SpSCC) was noted and confirmed by focal positivity of p63 in epithelial and sacromatoid components. The pleomorphic sarcomatoid tumor was positive for vimentin with Ki67 highlighting 70% of tumor cells. A final diagnosis of sinonasal spindle cell carcinoma associated with residual inverted papilloma was rendered.

Conclusions: Due to the rarity of such cases, the prognosis and response to treatment is unclear. No effective directed treatment has been developed. Unfortunately, the patient refused any further treatment and died of persistent disease. To the best of our knowledge, only one case of sinonasal carcinosarcoma arising from dysplastic inverted papilloma has been reported. The distinct possibility of previous radiotherapy contributing to development of sarcomatoid features in this neoplasm should also be considered.

MeSH Keywords: Carcinoma, Squamous Cell • Carcinosarcoma • Papilloma, Inverted

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/921827

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Background

Sarcomatoid carcinoma, also known as spindle cell carcinoma (SpCC), is an uncommon and distinctive form of squamous cell carcinoma (SCC) that bears both diagnostic and treatment challenges. SpCC is distinctive due to biphasic pattern of the tumor, namely, the poorly differentiated epithelial component of SCC and a spindle cell component. The larynx is the most common site in the aerodigestive tract. Other primary sites of SpCC, although less common, include the oropharynx, hypopharynx, and sinonasal tract [1,2]. Several cases of SpCC have been reported in the sinonasal area [2–4]. However, sinonasal SpCC in a setting of inverted papilloma has rarely been described in literature [4,5]. Herein, we present an unusual case of sinonasal SpCC with synchronous inverted papilloma developing in the left maxillary sinus and nasal cavity in a 72-year-old male with a history of radiotherapy for sinonasal SCC 3 decades earlier.

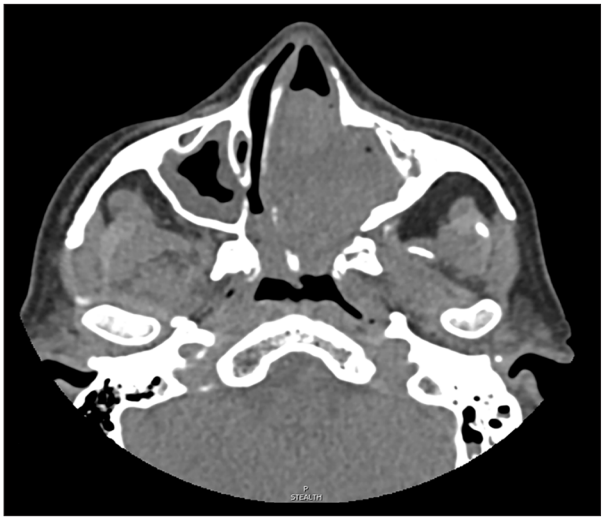

Figure 1. Axial section of computed tomography demonstrates a massive tumor occupying the left nasal cavity and maxillary sinus and destroying the nasal bones including bowing of the nasal septum on the affected side.

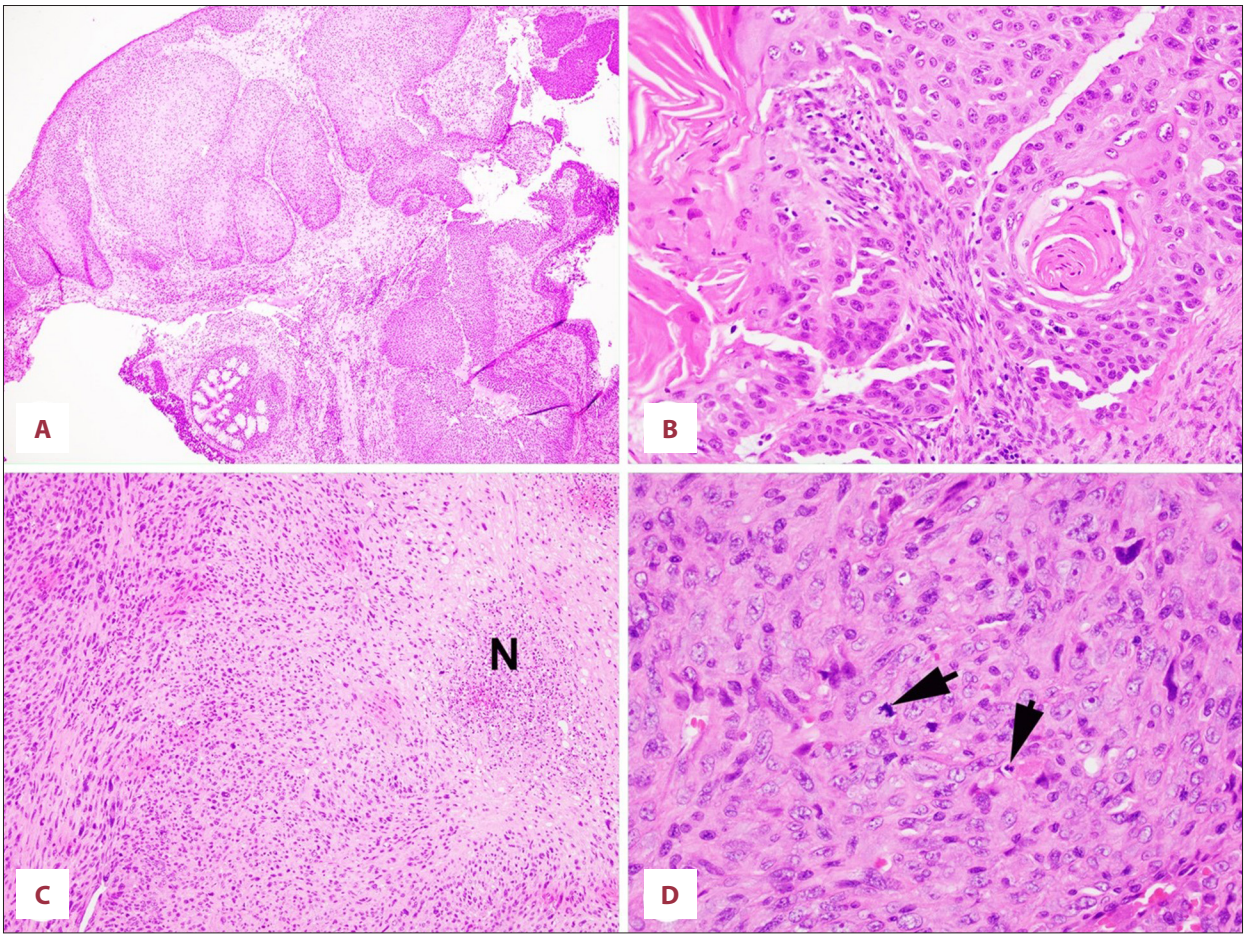

Figure 2. (**A**) Low power photomicrograph showing an inverted papilloma. (**B**) Medium power shows epithelioid neoplastic cells with keratin pearl formation. (**C**) Lower power magnification shows sarcomatoid areas with foci of necrosis. (**D**) High power of sarcomatoid area shows brisk mitosis (arrows), cellular and nuclear pleomorphism and hyperchromatism.

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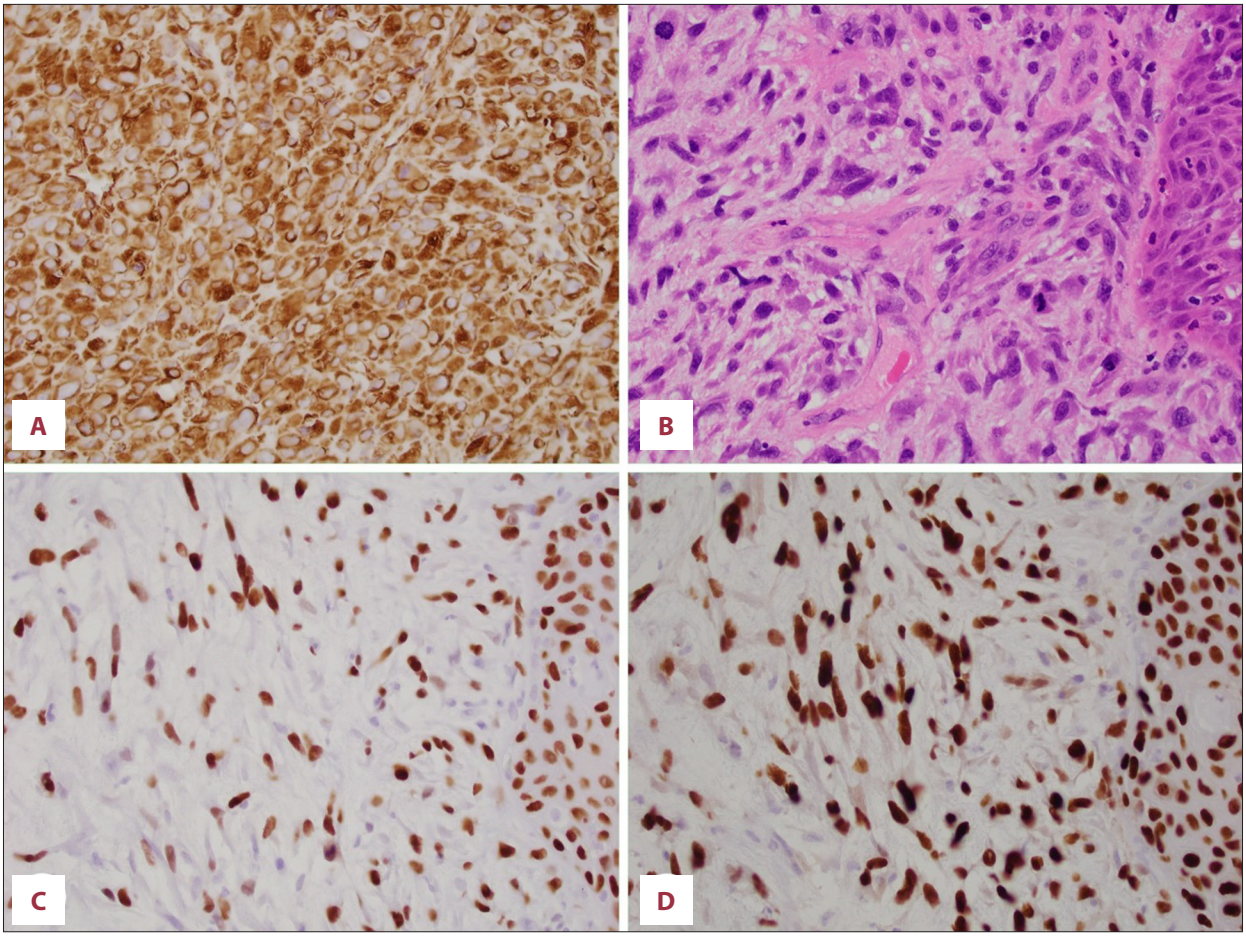

Figure 3. 400× magnification of (**A**) the sarcomatoid area demonstrating diffuse strong positivity for vimentin, (**B**) transitional area with sarcomatoid changes subjacent to the invading well differentiated islands, (**C**) strong positivity for p40 immunostaining in both the spindle cells and the adjacent intact atypical epithelium, and (**D**) strong diffuse positivity for p63 in the same tissue as photomicrograph C.

Case Report

A 72-year-old male was referred to our ENT (Ear, Nose and Throat) clinic with chief complain of left nasal obstruction with purulent nasal drainage, diminished sense of smell and intermittent episodes of epistaxis. The patient had a significant history of sinonasal SCC for which he underwent radiation therapy 30 years ago. The patient experienced symptoms of nasal obstruction a year ago and was seen by an otorhinolaryngologist. Further investigation yielded a diagnosis of biopsy-proven sinonasal polyp that had recurred after excision. The patient was referred to UF Health ENT clinic for further evaluation.

Nasal endoscopy revealed an ulcerated, friable, well-circumscribed mass occupying left nasal vestibule and left maxillary antrum. No palpable lymphadenopathy was identified.

Computerized tomography (CT) imaging demonstrated a lobular soft tissue mass that leads to tight nasopharynx and massive destruction of the palatal bone (Figure 1). The tumor was resected through endoscopic surgical procedure. Macroscopically, a white fleshy appearance with necrosis was noted in the submitted specimen. Microscopically, the tumor was composed of pleomorphic epithelial and spindle cells with numerous mitoses and remarkable tissue necrosis. Residual inverted papilloma (IP) with high-grade dysplasia, and minimal foci of moderately differentiated squamous cell carcinoma (SCC) component was present at the tumor margin. A distinct zone of transition of SCC to SpSCC was noted and confirmed by focal positivity of p63 in epithelial and sacromatoid components. The pleomorphic sarcomatoid tumor was positive for vimentin with Ki67 highlighting 70% of tumor cells (Figure 2). The pleomorphic sarcomatoid tumor was negative for the other epithelial markers (p40, CK5/6, AE1/AE3), neural crest makers (S-100 and Sox10), vascular markers (CD34, CD31, and ERG1), and myogenic markers (SMA, desmin, and myogenin) (Figure 3). A final diagnosis of sinonasal spindle cell carcinoma associated with residual IP was rendered. The patient succumbed to his disease a few months later.

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Discussion

SpCC is a specific type of SCC that displays pleomorphic or spindle cells with epithelial neoplastic cells. Like conventional SCC, it mostly shows a remarkable male predilection in the fifth and sixth decades of life and is strongly linked to smoking, alcohol consumption, and a history of radiotherapy [4,6]. This patient also had history of adjunctive radiotherapy for sinonasal SCC 30 years earlier. The sarcomatoid changes seen in the specimen could be attributed to this previous radiation therapy. Radiation to head and neck region can induce soft tissue sarcomas within a few months to several years with a median period of 7.35 years. These sarcomas, including carcinosarcoma are associated with overall unfavorable prognosis [7].

SpCC clinically present as a polypoid exophytic and ulcerated mass [1,6]. Our case showed similarity in clinical endoscopic presentation when compared with previous case reports. This clinical pattern, which is similar to other benign sinonasal lesions such as sinonasal polyps, can mistakenly indicate a benign clinical entity. In our case a diagnosis of sinonasal polyp was rendered before it was resected, and recurrence was noted.

Lio et al. found that there was no relationship between sinonasal inverted papilloma and SpCC. Nevertheless, the risk of malignant transformation of sinonasal IP to any variant of SCC, including SpCC, does not completely exclude the potential association between them [4].

It has been speculated that surgery with chemoradiation improves the patient prognosis [8]. However, high level of evidence is not available for the optimal management of SpCC due to the paucity of this entity [9]. Alem and Alnoury reported a 67% recurrence rate for sinonasal SpCC [8]. Other authors have reported a mortality rate of 42% in 2.5 years for SpCC in other locations in the body, which reflects the significant aggressive behavior of this tumor [10,11]. No definite conclusion for prognosis of SpCC of head and neck was noted in a case series analyzed and reported by Iqbal et al. However, they concluded that the tongue SpCC are more aggressive regardless the treatment modality used [9].

Conclusions

The SpCC is an unusual variant of SCC that can occur in setting of sinonasal inverted papilloma. It can be clinically and histologically challenging. The sarcomatoid changes may be linked to prior radiation therapy in the region. Most treatment in the literature is based on anecdotal reports and long-term clinical follow-up is essential to prevent future recurrences and to determine need for additional therapy.

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