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Case Report

Sinonasal teratocarcinosarcoma: a case report of a 41-year-old female

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Abstract: Sinonasal teratocarcinosarcoma (SNTCS) represents an exceedingly rare and aggressive malignancy, exhibiting a unique combination of teratomatous and carcinosarcomatous features. Phenotypically, these neoplasms consist of benign neural components alongside various malignant epithelial and mesenchymal elements. Consequently, the clinical presentation and therapeutic strategies for this tumor entity still necessitate further case-based elucidation. A case involving a 41-year-old female diagnosed with SNTCS is delineated herein. The patient had experienced right-sided nasal obstruction and intermittent epistaxis persisting for one month. Clinical examination identified a polypoid mass occupying the right nasal cavity, which was subsequently excised surgically. Histopathologic and immunohistochemical study revealed a heterogeneous admixture comprising adenocarcinoma, chondrosarcoma, primitive neuroendocrine elements, and immature squamous components of teratoma origin, Positive staining for epithelial membrane antigen and pancytokeratin was observed in both tumor and neuroepithelial cells. Vimentin expression was positive. Partial positivity was demonstrated for CD99, synaptophysin, chromogranin A, and S-100 protein. Focal positivity was identified for desmin and glial fibrillary acidic protein. Ki-67 staining was positive in approximately 60% of cells within the visual field, substantiating the diagnosis of SNTCS. Given its high degree of malignancy and local invasiveness, complete surgical resection and extensive tissue sampling remain imperative for an accurate diagnosis. Prompt identification and comprehensive management have been associated with improved prognostic outcome.

Keywords: Aggressive malignant tumor, pathologic examination, sinonasal teratocarcinosarcoma, surgery

Introduction

Sinonasal teratocarcinosarcoma (SNTCS) is a rare malignancy, comprising less than 5% of all head and neck tumors [1], and is histologically characterized by epithelial, mesenchymal, and neuroectodermal components [2]. This neoplasm most frequently arises within the nasal cavity, often infiltrating the ethmoid and maxillary sinuses [3]. It is distinguished by its high-grade malignancy and pronounced local invasiveness, which collectively contribute to an elevated recurrence rate. Literature indicates that its three-year survival rate does not exceed 40%, Identified risk factors include occupational exposure to wood dust and various industrial agents, tobacco consumption, and human papillomavirus infection [4]. Within the United States, the incidence is estimated at fewer than 1 per 100,000 individuals. Local expansion of the tumor may induce symptoms such as nasal congestion, obstruction, epistaxis, and anosmia, whereas progression into contiguous structures, including the orbit, oral cavity, nasopharynx, and skull base, can result in visual disturbances and alterations in facial morphology [5]. Owing to the rarity of SNTCS, this entity is frequently misdiagnosed as other sinonasal tumors, including olfactory neuroblastoma, small cell carcinoma, immature teratoma, teratoma with malignant transformation, carcinosarcoma, or adenocarcinoma [6]. Historical diagnostic inaccuracies have contributed to limited understanding, with fewer than 150 cases documented in existing medical literature. A marked male predominance has been observed, with a reported male-to-female ratio of 7:1 and a mean age of onset of approximate-

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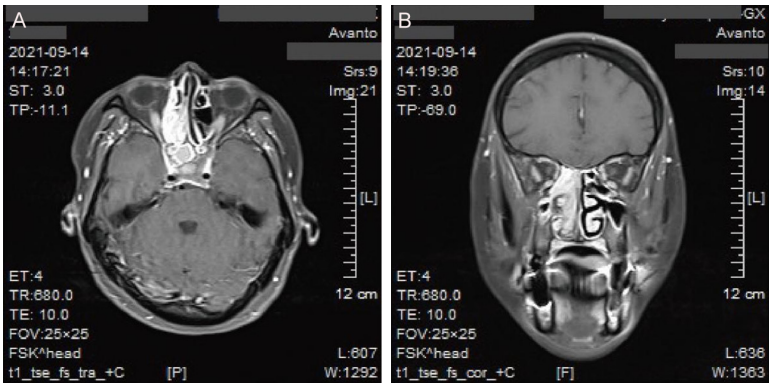

Figure 1. Simultaneous enhanced magnetic resonance imaging (MRI) of the paranasal sinuses T1:TR: 498.0, TE: 10.0. T2:TR: 6030.0, TE: 88.0. A: Right nasal cavity and paranasal sinus mass lesion with long T1 and long T2 signal intensity. The lesion exhibits heterogeneous internal signals with scattered small patchy areas of short TI signal foci. B: After contrast enhancement, the lesion shows marked heterogeneous enhancement. Bilateral ethmoid sinuses, sphenoid sinuses, and maxillary sinuses demonstrate long T1 and long T2 signal enhancement. Additionally, a T1 signal is visible within the right sphenoid sinus.

ly 54.7 years. Misra et al. conducted a review encompassing 86 published cases of SNTCS, identifying nasal obstruction as the most prevalent presenting symptom, followed by epistaxis. Additional reported manifestations included headache, visual impairment, and olfactory dysfunction [7]. In this study, a case involving a 41-year-old female diagnosed with SNTCS is reported. Total excision and extensive sampling are necessary to reach the diagnosis. Early diagnosis and management can give a better prognosis.

Case information

Clinical history

A 41-year-old female was admitted with a primary complaint of right nasal swelling and right-sided nasal obstruction persisting for one month. Pre-admission electronic nasopharyngoscopy revealed a dark red neoplastic mass located in the right nasal cavity, possessing a pedicle that appeared to originate from the olfactory cleft and extending posteriorly toward the nasopharynx. Dark red secretions were identified in both the right common nasal meatus and the middle nasal meatus, while the mucosa of the nasopharynx appeared smooth. The patient was referred to this institution for further diagnostic assessment and therapeutic intervention. Upon physical examination, a smooth-surfaced, erythematous mass was observed in the right nasal cavity, prone to spontaneous bleeding. Bilateral hypertrophy of the middle and inferior turbinates was noted, accompanied by an "S"-shaped deviation of the nasal septum, absence of paranasal sinus tenderness, and reduced olfactory acuity. A clinical diagnosis of a right nasal cavity mass was established. Further evaluation with paranasal sinus computed tomography (CT) and magnetic resonance imaging (MRI) was recommended.

Imaging studies

Sinus CT (**Figure 4A**, **4B**) revealed soft tissue opacification within the right nasal cavity and nasopharynx, accompanied by morphologic enlargement of the ostiomeatal complex and obstruction of the sinus ostium. Soft tissue density shadows were detected within the bilateral ethmoid, sphenoid, and maxillary sinuses, alongside diminished aeration of the affected sinus cavities. Moreover, no evident abnormalities were identified in the bony structures of the sinus walls.

MRI of the paranasal sinuses (**Figure 1A**, **1B**) demonstrated a space-occupying lesion in the right nasal cavity and paranasal sinuses. After contrast enhancement, the lesion exhibited marked heterogeneous enhancement. Long T1 and long T2 signal intensities with enhancement are observed within the ethmoid sinuses, sphenoid sinuses, and maxillary sinuses. No tumor invasion of the base of the skull was observed.

Chest CT exhibited no apparent abnormalities.

Treatment

Following a comprehensive evaluation, endoscopic resection of the right nasal cavity and paranasal sinus tumor, in conjunction with right sinusotomy, right middle turbinate reconstruction, and fracture with lateral displacement of the right inferior turbinate, was recommended. Under general anesthesia, complete excision of the right nasal cavity tumor was performed, measuring approximately 2.3 cm × 1.0 cm ×

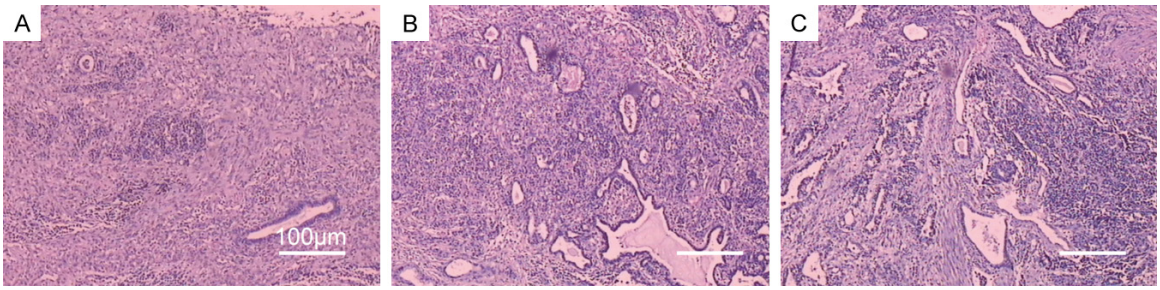

Figure 2. A: The cancerous tissues and poorly differentiated sarcoma areas are mostly accompanied by fibrous connective tissue hyperplasia, with focal evidence of primitive neuroectodermal differentiation. B: Predominance of cancerous tissue and poorly differentiated sarcoma regions, with fibrous tissue hyperplasia. C: Cancerous tissue and poorly differentiated sarcoma areas, accompanied by fibrous tissue hyperplasia, and focal smooth muscle. Bar: 100 μm, 40X.

0.5 cm. The procedure was executed successfully, and no postoperative complications were encountered. A combination of antibiotics and hemostatic agents was administered for three days following surgery. Additionally, no residual tumor was detected within the nasal cavity, indicating a favorable postoperative recovery.

Pathologic examination

Following excision of the polypoid mass, hematoxylin and eosin staining revealed a malignant neoplasm exhibiting a variegated histologic appearance. The tumor was composed of glandular epithelial elements and spindle-shaped mesenchymal components, accompanied by areas of inflammatory necrosis. Carcinomatous, sarcomatous, primitive neuroectodermal, and teratomatous components were found to be intricately intermixed. Additionally, multifocal poorly differentiated sarcomatoid regions were identified surrounding adenocarcinoma and mature brain tissue, along with areas containing smooth muscle and tumor-like proliferative fibrous tissue (**Figure 2A-C**).

Immunohistochemical analysis demonstrated positive staining for epithelial membrane antigen (EMA) and pancytokeratin (CKpan) in both tumor and neuroepithelial cells (**Figure 3A** and **3B**). Vimentin expression was also positive (**Figure 3C**). Partial positivity was observed for CD99, synaptophysin (Syn), chromogranin A (CgA), and S-100 protein (**Figure 3D-G**). Desmin and glial fibrillary acidic protein (GFAP) showed focal positivity (**Figure 3H**, **3I**). Approximately 60% of the visual field exhibited Ki-67 positivity (**Figure 3J**).

Diagnosis and treatment

The diagnosis was revised to teratocarcinosarcoma of the right nasal cavity and paranasal sinuses. Due to its high malignancy, approximately 60% of patients survive for no more than 3 years. Although complete surgical excision had been achieved, due to its extensive lesion range, tumor cells may have remained in areas that were difficult to completely remove through surgery. Furthermore, influenced by the local microenvironment, these residual cells are prone to re-proliferation, leading to disease recurrence. Additionally, a larger lesion range often indicates a higher risk of tumor cell infiltration and metastasis. Postoperative radiotherapy can deliver precise irradiation to the primary lesion and potential subclinical lesions, effectively killing residual cancer cells and reducing the local recurrence rate and the risk of distant metastasis. Radical surgery in combination with postoperative radiotherapy was still recommended. The patient was advised to seek further consultation and advanced treatment at a tertiary care center. Subsequent follow-up was not achieved, as the patient was lost to follow-up. Despite multiple attempts to reestablish contact through phone calls, text messages, and mailed reminders, there was no response from the patient or their designated caregiver. The patient's last known address and contact information, which had been provided during initial registration, proved to be outdated or incorrect, leading to undelivered mail and unanswered calls. Attempts to reach out by alternative means, such as checking local community health centers or contacting family members

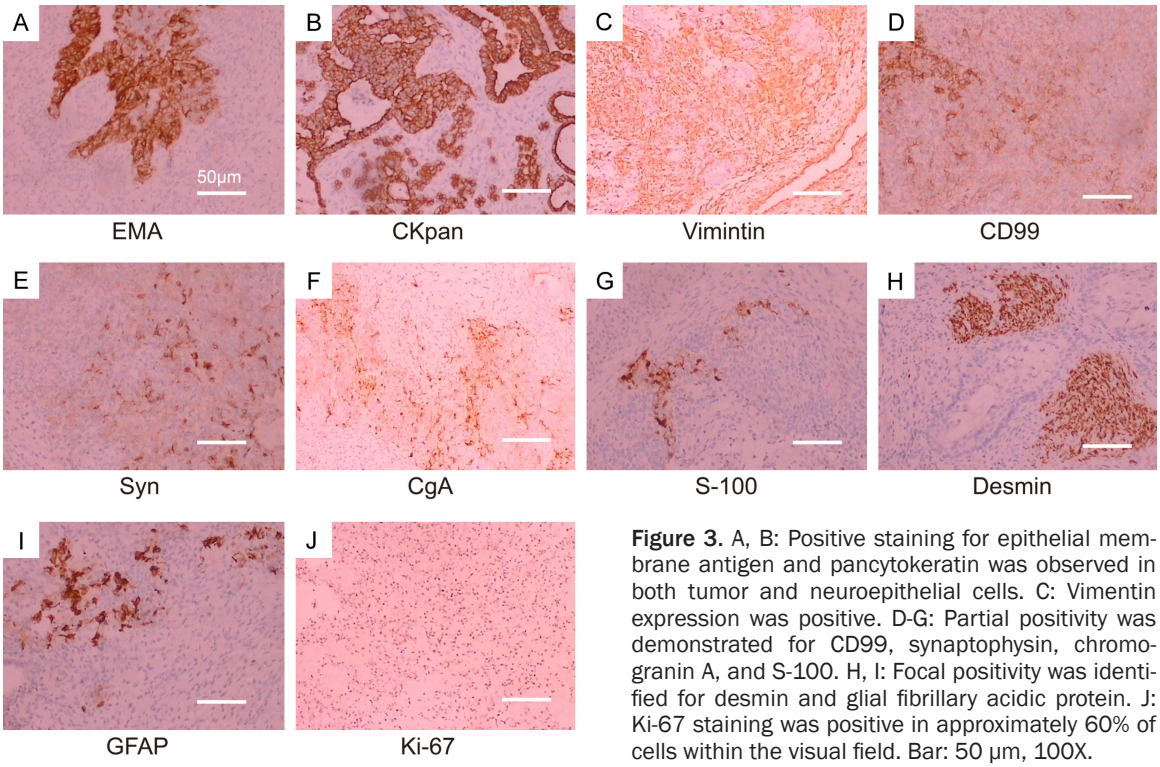

Figure 3. A, B: Positive staining for epithelial membrane antigen and pancytokeratin was observed in both tumor and neuroepithelial cells. C: Vimentin expression was positive. D-G: Partial positivity was demonstrated for CD99, synaptophysin, chromogranin A, and S-100. H, I: Focal positivity was identified for desmin and glial fibrillary acidic protein. J: Ki-67 staining was positive in approximately 60% of cells within the visual field. Bar: 50 μm, 100X.

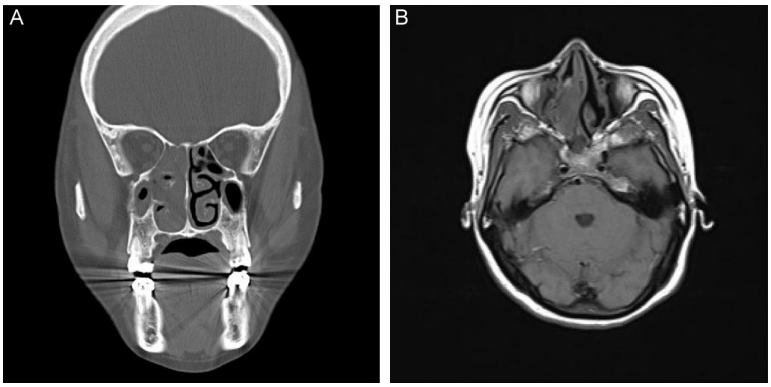

Figure 4. A: Coronal computed tomography (CT) of the Paranasal Sinuses. Soft tissue density shadow in the right nasal cavity and paranasal sinuses, with enlargement and obstruction of the nasal sinus ostium complex. B: Axial CT of the paranasal sinuses shows soft tissue density shadows in the right nasal cavity and nasopharynx, which blocks the airway. No obvious abnormalities are observed in the bony structures of the sinus walls.

listed in the patient's records, were also unsuccessful, as those contacts were either unreachable or unwilling to provide updated information. The loss to follow-up resulted in an inability to monitor the patient's progress, administer necessary post-treatment care, or address any complications that may have arisen in the interim period.

Discussion

SNTCS is a highly aggressive and rare malignancy characterized by a heterogeneous composition of malignant blastematous, epithelial, and mesenchymal components. Its precise histogenetic origin remains undefined [5]. The global incidence of SNTCS is exceedingly low, with only approximately one hundred cases documented to date [8]. The nasal cavity and paranasal sinuses represent the most frequently affected anatomical sites, followed by the nasopharynx, lateral pharyngeal wall, and anterior skull base [9]. Clinically, patients often present with nasal obstruction and epistaxis, and may also experience anosmia, headache, visual disturbances, and neurological deficits [6]. In the present case, the patient presented with nasal obstruction of one month's duration, and nasal endoscopy at admission revealed a red neoplasm

Table 1. Significance of key positive markers

Landmark	Positive site	Pathologic significance	Diagnostic Prompt Direction
EMA	Tumor cells/ neuroepithelium	Epithelial tumor classic marker, which can be positive in neurogenic tumors (such as ependymomas)	Origin from epithelium or special neuroepithelium
CK-pan	Tumor cells/ neuroepithelium	Broad-spectrum epithelial marker; positivity in non-epithelial tumors (such as synovial sarcoma, some neuroendocrine cancers) requires vigilance	Epithelial differentiation or biphasic differentiated tumor
Vimentin	Tumor cells/ neuroepithelium	Mesenchymal lineage markers, coexistence with EMA/CKpan suggests - Part of carcinoma/sarcoma - Sarcomatoid Carcinoma - Ependymomas and other neuroepithelial tumors	The tumor exhibits an epithelial-mesenchymal biphenotype
CD99	Some tumor cells	Common in: - Solitary Fibrous Tumor - Solitary Fibrous Tumor - Part of carcinoma/sarcoma	Need to combine morphology to rule out PNET family tumors
Syn/CgA	Some tumor cells	Neuroendocrine differentiation markers (Syn is more sensitive, CgA has high specificity)	Neuroendocrine tumors (such as paragangliomas and neuroendocrine carcinomas)
S-100	Some tumor cells	Neural sheath/melanocyte/adipose-derived tumor markers, which can be focally positive in neuroendocrine tumors	Supports neuroendocrine or neuroendocrine differentiation
Desmin	Focal positivity	Myogenic differentiation markers (rhabdomyosarcoma/smooth muscle sarcoma), focal positivity may indicate abnormal expression	Sarcoma components need to be excluded
GFAP	Focal positivity	Glial cell markers (astrocytoma/ependymoma) are rare in neuroendocrine tumors	Suggests possible neuroepithelial differentiation
Ki-67 (60%)	Tumor cells	High proliferative activity (with >20% indicating high proliferation) is consistent with the characteristics of high-grade malignant tumors	Aggressive in nature, with a poor prognosis

Abbreviations: EMA: epithelial membrane antigen; CKpan: pan cytokeratin; Syn: synaptophysin; CgA: chromogranin A; GFAP: glial fibrillary acidic protein.

consistent with the initial diagnostic characteristics of SNTCS.

SNTCS typically lacks distinctive CT or MRI features, rendering differentiation from other common malignant sinonasal neoplasms challenging [10]. In the present case, CT imaging revealed nonspecific tumor characteristics. SNTCS frequently demonstrates considerable histological heterogeneity, and definitive pathological and immunohistochemical confirmation can be achieved only through thorough examination of the entire resected specimen. The tumor is generally characterized as a heterogeneous composition comprising elements derived from all three germ layers, exhibiting variable degrees of differentiation. Prior studies have indicated that immunohistochemical analysis contributes markedly to the diagnostic process for SNTCS [9-11]. The staining profile is largely determined by cellular subtype, including positive staining for epithelial markers (cytokeratin and EMA), mesenchymal markers (vimentin), and undifferentiated or primitive tumor components (CD99 and NSE). Additionally, positive immunoreactivity is often observed in neuroepithelial elements, including neuron-specific enolase, CD99, CgA, Syn, GFAP, and S-100 protein, with Syn and CgA frequently demonstrating weak positivity. These staining characteristics were largely concordant with the immunohistochemical findings in the present case. Notably, although some reports describe negative staining for SMA and desmin, other investigations have documented reactivity to myogenic markers, including smooth muscle actin [11]. In the current case, focal positivity for desmin was observed on immunohistochemical evaluation. Meanwhile, the significance and comparison of some key markers are shown in **Table 1**.

Because of the rarity of SNTCS, a consensus regarding its optimal management has not yet been established. Current treatment strategies are largely informed by limited case series and retrospective analyses [12]. Multimodal therapeutic approaches, typically involving surgery followed by radiotherapy and/or chemotherapy, are considered necessary to optimize oncological outcomes. Nevertheless, the overall prognosis remains poor, with substantial risks of both metastasis and local recurrence. The mean 2-year survival rate has been reported at 55%, with a recurrence rate of 38% [13]. Neoadjuvant therapy, whether followed by surgery or administered in conjunction with radical chemoradiotherapy, has been proposed as a promising therapeutic strategy. Management of SNTCS generally incorporates surgery, radiotherapy, and chemotherapy, with various combinations employed to achieve maximum benefit [5]. The most frequently chosen treatment modalities include surgical resection (87.2%), radiotherapy (59.3%), and chemotherapy (18.6%). Surgical intervention with negative margins remains the preferred method [6]. In the present case, the patient was lost to follow-up after discharge, precluding determination of whether the recommended treatment at the tertiary care facility was ultimately pursued.

Conclusions

SNTCS represents a rare and highly invasive tumor entity of the sinonasal region, associated with poor prognosis. Improved clinical outcomes may be achieved through early diagnosis followed by complete surgical resection with thorough histopathologic sampling, in combination with intensive adjuvant therapy.

Disclosure of conflict of interest

None.

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