Clinical Case Report

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Local recurrence of sinonasal renal cell-like adenocarcinoma

A CARE compliant case report

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Abstract

Rationale: Sinonasal renal cell-like adenocarcinoma (SNRCLA) is a very rare sino-nasal carcinoma. Because SNRCLA has the same morphological features as other clear cell carcinomas, and some of them also occurred in sinonasal part, it is necessary to differentiate SNRCLA from these tumors.

Patient concerns: A 42-year-old man presented with complaints of epistaxis for 1 day. The patient had undergone endoscopic resection of a neoplasm in the right nasal passage at another hospital 35 months before and was diagnosed with SNRCLA at that time, and did not receive any other adjuvant therapy.

Diagnoses: The postoperative histopathological examination revealed a diagnosis of recurrent SNRCLA.

Interventions: The tumor was removed under nasal endoscopy.

Outcomes: The patient was followed up for 2 months and recovered well without any complications.

Lessons: NSRCLA is a very rare tumor, and should be differentiated from other clear cell tumors including some salivary tumors and metastatic tumors of renal and thyroid.

Abbreviations: ACC = acinic cell carcinoma, CCFTC = clear cell variant follicular thyroid carcinoma, CCPRCC = clear cell papillary variant renal cell carcinoma, CCRCC = clear cell variant renal cell carcinoma, EMC = epithelial–myoepithelial carcinoma, RCC = renal cell carcinoma marker, SCCC = salivary clear cell carcinoma, SNRCLA = sinonasal renal cell-like adenocarcinoma.

Keywords: differentiation, metastatic CCRCC, recurrent, SCCC, SNRCLA

1. Introduction

Sinonasal renal cell-like adenocarcinoma (SNRCLA) is a very rare tumor that occurs in the sinonasal part because that there are only about 20 reported cases in English literature since the first 2 SNRCLA cases were reported in 2002.[1,2] SNRCLA has the microscopic features similar to some metastatic sinonasal carcinomas including clear cell renal cell carcinoma (CCRCC) and follicular thyroid carcinoma.[3–5]

The morphology of SNRCLA was so mild that people once questioned whether it was suitable to name it adenocarcinoma instead of benign adenoma.[6] However, in spite of most patients with SNRCLA received adjuvant radiotherapy after surgery, accumulating data revealed that about 20% patients with SNRCLA still had tumor recurrence.[6,7] Here, we reported a case of recurrent SNRCLA in the right ethmoid sinus 35 months after tumor removal. This study was approved by the Ethical Committee of Second Military Medical University Gongli Hospital (No. 2018-75). Informed written consent was obtained from the patient for publication of this case report and accompanying images.

2. Case presentation

A 42-year-old man presented with complaints of epistaxis for 1 day, and computed tomography scan showed a soft tissue mass with increased density in the right ethmoid sinus (Fig. 1). In the past, the patient had undergone endoscopic resection of a neoplasm in the right nasal at another hospital 35 months ago. The patient was diagnosed with SNRCLA at that time and did not receive any other treatments such as chemoradiotherapy after surgery. According to the patient's history, we considered the patient as the recurrence of SNRCLA. Under general anesthesia, the nasal mucosa surface was conversed with 0.11% adrenalin, and the neoplasm in the right ethmoid sinus was removed.

Macroscopically, the tumor was composed of a pile of gray–white, broken tissue measuring 1.5 cm in diameter. The tumor was mainly composed of clear cells, which might be mildly acidophilic, and had small blood vessels in the tumor stroma (Fig. 2A). The clear cells were arranged with solid, tubular and follicular structure, and eosinophilic secretions were visible in the lumen, similar to that of thyroid tissue (Fig. 2B). The heterogeneity of clear cells was very small due to mild nucleus and rare mitosis (Fig. 2C). Hemorrhage of tumor stromal tissue

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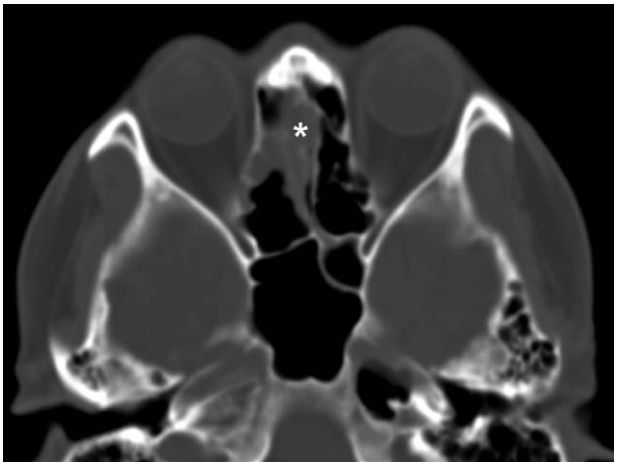

Figure 1. CT scan revealed a soft tissue mass with increased density in the right ethmoid sinus (A, as showed by the asterisk). CT = computed tomography.

was obvious in focal areas, like that of CCRCC (Fig. 2D). The tumor cells were positive for CAX9 (Fig. 3A), CK7 (Fig. 3B), S100 (Fig. 3C), SOX10 (Fig. 3D), Vimentin (Fig. 3E), and EMA, whereas negative for CD10, P63, TTF-1, and Pax-8. The index of Ki67 was about 2% (Fig. 3F). According to both morphological and immunohistochemical features, the diagnosis of SNRCLA recurrence was made. The patient recovered well and was discharged on the fourth postoperative day without any complications. The patient was followed up for 2 months and recovered well without any complications.

3. Discussion

SNRCLA is a rare, low-grade neoplasm that bears no resemblance to any other sinonasal primary tumor.[8] The tumor has a female predominance of 2:1. The clinical manifestations are relatively nonspecific including epistaxis, nasal obstruction, headache, and olfactory impairment. The nasal cavity is the most common site of involvement, followed by paranasal sinuses and nasopharynx.

As showed in Table 1, SNRCLA should be differentiated from other clear cell tumors due to the similar morphological features, including salivary tumors and metastatic clear cell carcinomas or clear cell variant thyroid follicular carcinoma. Salivary of clear cell carcinomas (SCCCs) mostly occurred in the oral cavity with the clinical presentation of swelling and mucous ulcer. SCCCs were usually smaller than 3 cm in size with an infiltrative growth pattern.[9] Monomorphous polygonal clear cells and stromal

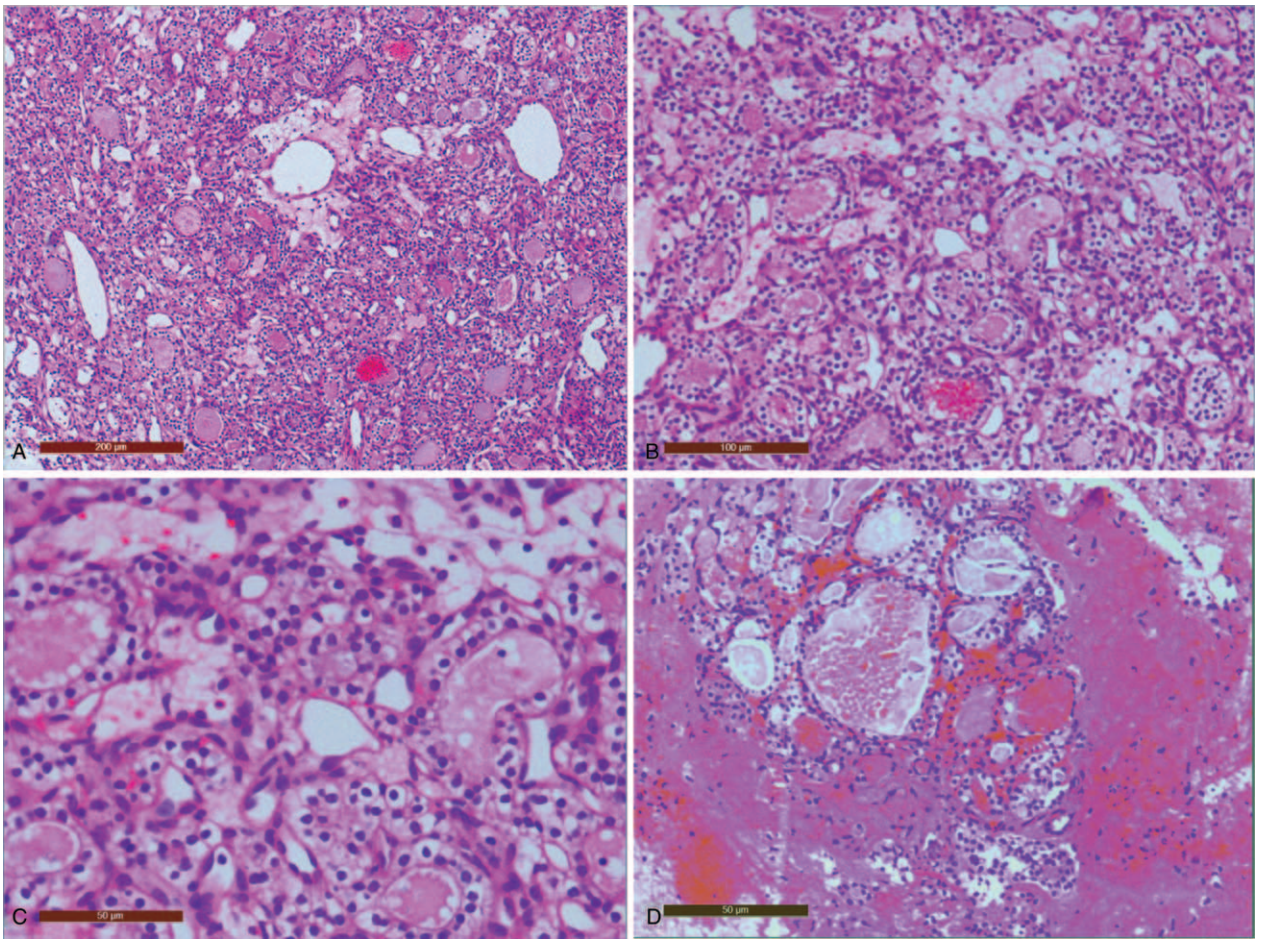

Figure 2. Histopathological features of SNRCLA. The tumor was mainly composed of clear cells, which might be mildly acidophilic, and had small blood vessels in the tumor stroma (A). The clear cells were arranged with solid, tubular and follicular structure, and eosinophilic secretions were visible in the lumen, similar to that of thyroid tissue (B). The heterogeneity of clear cells was very small due to mild nucleus and rare mitosis (C). Hemorrhage of tumor stromal tissue was obvious in focal areas, like that of CCRCC (D). CCRCC = clear cell variant renal cell carcinoma, SNRCLA = sinonasal renal cell-like adenocarcinoma.

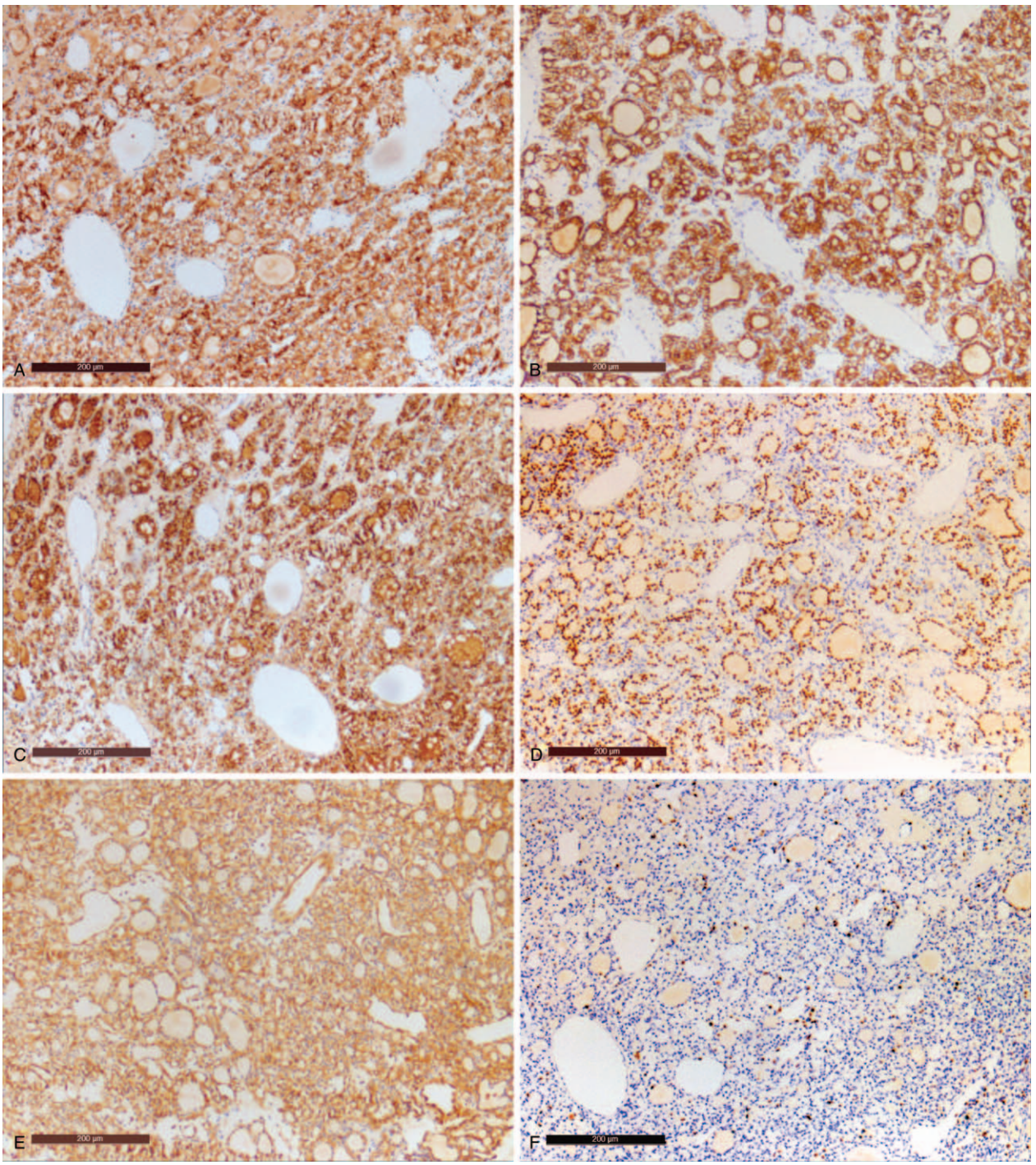

Figure 3. Immunohistochemical features of SNRCLA. Tumor cells were positive for CAX9 (A), CK7 (B), S100 (C), SOX10 (D), and Vimentin (E), and the index of Ki67 was about 2% (F). SNRCLA = sinonasal renal cell-like adenocarcinoma.

Table 1

Recurrent cases of SNRCLA.

Author	Age, Sex	Tumor site	Treatment	Follow up
Heffner[7]	62, F	Nasal	Surgery, adjuvant radiotherapy	Recurrence at 3 and 5 yr
Shen[6]	56, F	Nasal, skull base	Surgery, adjuvant radiotherapy	Recurrence at 22 mo
Shen[6]	89, F	Sinonasal	Surgery	Recurrence at 4 mo
Shen[6]	73, M	Nasal	Surgery, adjuvant radiotherapy	Recurrence at 20 mo
Present case	42, M	Sinonasal	Surgery	Recurrence at 35 mo

SNRCLA = sinonasal renal cell-like adenocarcinoma.

Table 2

The differentiation between SNRCLA and other clear cell tumors.

	Salivary tumors			RCC			
Tumors' name	**SCCC**	**EMC**	**ACC**	**Metastatic CCRCC**	**CCPRCC**	**CCFTC**	**SNRCLA**
Localization	Oral cavity	Parotid, minor gland	Parotid	Head and neck metastases, not nasal metastases	No lymph node or other metastases	Metastases of head and neck, not sinus	Sinus, nasal cavity
Presentation	Swelling, ulcer	Painless, slow-growing mass	Slow-growing mass	Hematuria, renal pain	Nonspecific, good prognosis	Thyroid mass	Hematuria, nasal obstruction
Gross features	<3 cm, Infiltrating growth	Multinodular mass, expansive borders	1–3 cm, Circumscribed nodules,	Bleeding, necrosis, clear border	Mass with multicolor and clear boundary	Round solid tumor with envelope	Polypoid or submucosal mass
Microscopic features	Monomorphous polygonal clear cell, stromal hyalinization	Spindled cells, basement membrane deposition.	Large, polygonal cells, lymphoid infiltration	Solid arrangement, nuclear heterogeneity	Papilla arrangement of eosinophilic cells	Follicular structure with clear or sigmoid cells	Monomorphous cuboidal glycogen-rich cells lacking heterogeneity
Immunohistochemical features	CK+, S100 −, CEA−	P63+, SMA+, CK+	PSA+, CK+, α 1-antitrypsin+,	Vimentin+, RCC+, CD10+	CK7+, RCC+, Vimentin +	TTF-1+, CK19+, Galectin+	CAX9+, CK7+, S100+

ACC = acinic cell carcinoma, CCFTC = clear cell variant follicular thyroid carcinoma, CCPRCC = clear cell papillary variant renal cell carcinoma, CCRCC = clear cell variant renal cell carcinoma, EMC = epithelial–myoepithelial carcinoma, RCC = renal cell carcinoma marker, SCCC = salivary clear cell carcinoma, SNRCLA = sinonasal renal cell-like adenocarcinoma.

hyalinization were the main morphological characteristics of SCCC. The tumor cells were positive for CK, but negative for S100 and CEA. Therefore, the sites of sinus and nasal cavity, no stromal hyaline degeneration and positive expression of S100 could help SNRCLA to rule out SCCC. Epithelial–myoepithelial carcinomas (EMCs) occurred mainly in parotid with the microscopic features of spindled cells and basement membrane deposition. The expression of myoepithelial marker P63/SMA could help the diagnosis of EMC.[10] Acinic cell carcinomas (ACCs) also occurred in parotid with slow-growing mass. The large, polygonal tumor cells and the stromal infiltration of lymphoid were morphological characteristics of ACC. Tumor cells had the positive expression of PSA and α1-antitrypsin, which were negative for the tumor cells of SNRCLA.[11]

The most important mimic of SNRCLA was metastatic CCRCC because both tumors had similar clear cells and a common background of hemorrhage. However, the nuclei heterogeneity and tumor necrosis of CCRCC were obvious. Tumor cells of CCRCC expressed positively CD10 and renal cell carcinoma marker (RCC), whereas SNRCLAs did not express these markers. In addition, clear cell papillary renal cell carcinoma (CCPRCC) also diffusely expressed CK7 and CAI9 like that of SNRCLAs, but CCPRCC is a low-grade, indolent neoplasm with not any report of the distant metastasis. CCPRCC expressed GATA3, which was negative for SNRCLA cells.[12] SNRCLA also should be differentiated from clear cell variant thyroid follicular carcinomas (CCTFC) because that both tumors had similar follicular structures composed of clear cells and eosinophilic secretions in the lumens. The tumor cells of CCTFC were positive for CK19 and TTF-1, which were negative for SNRCLA cells.[5]

SNRCLA was considered as a low-grade malignancy due to mild morphological features. Although no metastatic case of SNRCLA had been reported, there were approximately 20% patients, who had a local recurrence. We summarized all the recurrent SNRCLA cases in English literature at Table 2. Recurrent patients included 3 female and 2 male patients. For the 17 patients with follow-up data, 33% (3/9) patients with postoperative radiotherapy relapsed whereas only 25% (2/8) patients without postoperative radiotherapy relapsed.[6,7] Therefore, we could infer that radiotherapy seemed not to work on the treatment of NSRCLAs.

4. Conclusions

NSRCLA is a very rare tumor, and we should differentiate it from other clear cell tumors including some salivary tumors, metastatic CCRCC/CCPRCC, and metastatic CCTFC.

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Visualization: Yu-Kai Qi.
Writing – original draft: Jian Wu.
Writing – review and editing: .
Writing – review and editing: Wei-Xiang Chen, Jian Wu.

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