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Glomus Tumor of the Sinonasal Tract
– Two Case Reports and a Review of Literature –

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Herein we describe two cases of nasal glomus tumor. Histological findings were typical, save for one which was quite large (3.1 cm in its greatest dimension) with an invasive growth pattern and increased ki-67 labeling index (up to 10%). These features raised a red flag of similarity to a recently described "invasive glomus tumor of nasal cavity", suggesting a more aggressive form of glomus tumor. However, objective criteria for this possibility is lacking at present and more similar case studies are needed to establish a truly aggressive form of glomus tumor.

Key Words : Glomus tumor; Nasal cavity; Paranasal sinuses

Glomus tumors are rarely found in the head and neck. These tumors are usually originated from the glomus body, which is a neuro-arterial structure that helps regulate the body temperature, rich in the distal extremity. To our knowledge, only 23 cases occurring in the sinonasal tract have been reported thus far, including one case report from Korea.[1] Literature review reveals its preponderance for the slight elderly (average 54), female gender, location of the nasal septum, size less than 1 cm, and chief complaints of epistaxis and/or nasal obstruction. We recently experienced 2 glomus tumors in the nasal cavity, one of which showed an invasive nature.

CASE REPORTS

Case 1

A 66-year-old woman presented with right-sided epistaxis and partial obstruction, with a duration of one month. A dark purple hemorrhagic nasal mass was observed in the right superoposterior nasal cavity. Computed tomography demonstrated a well-enhancing 3.1-cm sized soft tissue mass with extension to the right sphenoid sinus through the sphenoethmoidal recess (Fig. 1). There was evidence of erosion of the medial wall of the left maxillary sinus and the ethmoid roof. The mass was excised, and pathological sections showed anastomosing trabeculae of uniform cells with a fibrolipomyxoid stroma surrounding a cavernous vascular network (Fig. 2). The cells had sharply punched-out rounded nuclei and faintly eosinophilic cytoplasm, indicating a glomus tumour. The immunoreaction of tumor cells, positive for alpha-smooth muscle actin and negative for CD34, supported the diagnosis (Fig. 3). The ki-67 labeling index was focally increased up to 10% (Fig. 4), but unequivocal histological evidences of malignancy such as atypical mitosis or vascular invasion were lacking.

Case 2

A 92 year old man presented with a 2-week history of epistaxis. Two dark purple hemorrhagic nasal masses were found, in the

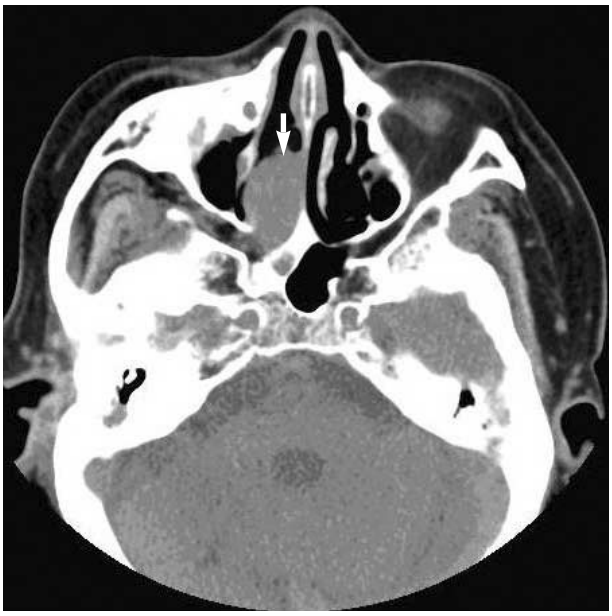

Fig. 1. A well-enhancing soft tissue mass at the right superoposterior nasal cavity is shown with extension to right sphenoid sinus, through sphenoethmoidal recess (arrow).

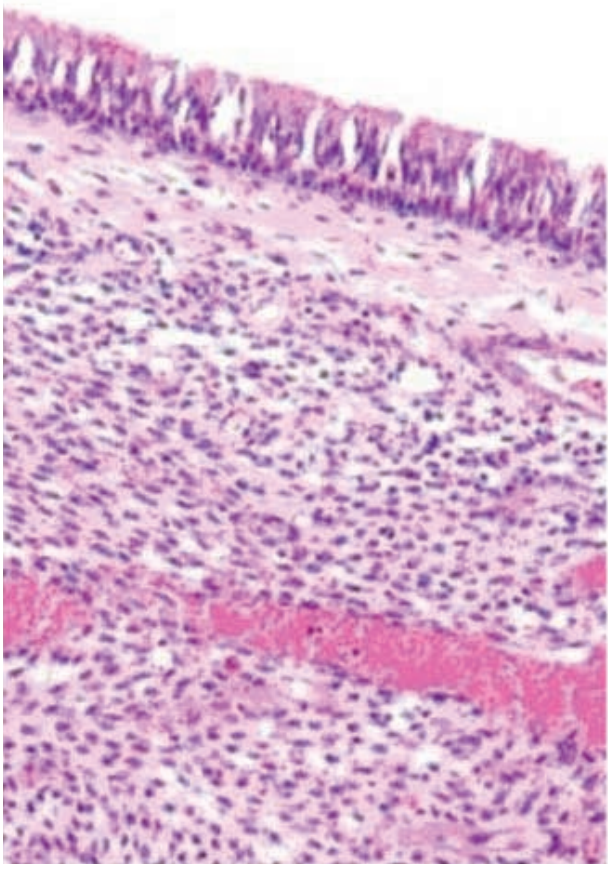

Fig. 2. The tumor cells are forming subepithelial sheets with indistinct cell borders. The ovoid or polyhedral cells with faintly eosinophilic cytoplasm are interrupted by vessels.

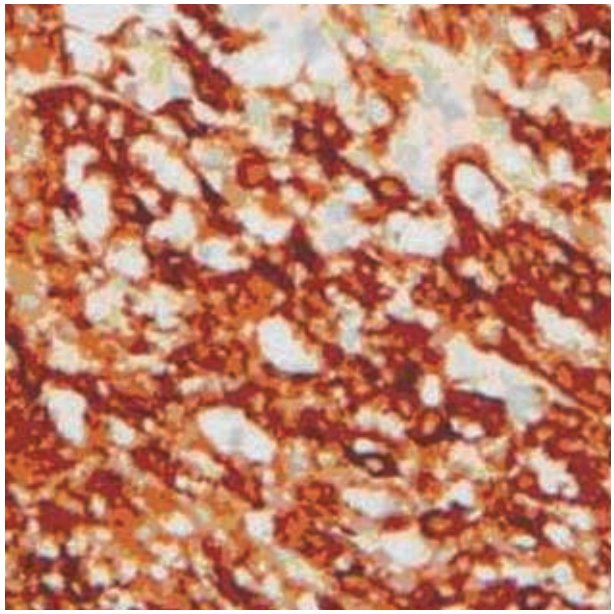

Fig. 3. The tumor shows diffuse immunopositivity for smooth muscle actin.

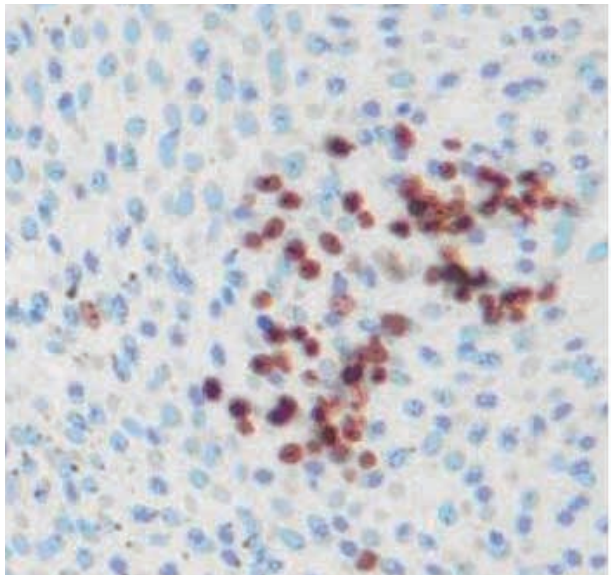

Fig. 4. The tumor shows focally increased ki-67 labeling index (up to 10%).

right posterior nasal septum (0.9 cm) and in the left medial side (0.6 cm). Image work-up was not performed, and by the nasal endoscopy, the right posterior nasal septum was eroded. Pathological findings were typical of a glomus tumor.

Both of our patients were successfully treated with endoscopic endonasal resection. At the time of this writing, they are alive and asymptomatic.

DISCUSSION

Glomus tumors are benign lesions, believed to represent hyperplasia or hamartomatous development of the glomus body.

The glomus bodies are most numerous in distal sites such as the digits, and glomus tumors are most frequently seen in the subungual area of the fingers. Occasionally they occur in the deep soft tissues, bone, vagina, trachea, lung, stomach, other gastrointestinal tract and rarely in the head and neck regions. Its variants are glomangioma and glomangiomyoma. Glomangioma resembles cavernous hemangioma due to glomuvenous malformation. Glomangiomyoma shows variable proportions of glomus cells, vascular structures, and smooth muscle cells.

Glomus tumors of the nose and paranasal sinus are extremely rare (Table 1). Most case reports make mention of an older population as being most often affected (average age, 54 years), though glomus tumors in the nasal cavity of young individuals have been also reported.[2-5] The male : female ratio was 1 : 1.9. According to the reported cases, obstruction and epistaxis are common presenting symptoms, and pain and asymptomatic presentation occur less frequently. Both of our patients presented with epistaxis, probably attributed to the vascularity of the tumors.

Although glomangiomas are benign, their rate of local recurrence is reported to be about 8%.[4,6] Most nasal glomus tumors have shown more indolent behavior, however, Hayes *et al*.[4] mentioned the repeated recurrences of a histologically benign sinonasal glomus tumor, without invasiveness or metastasis. Gaut *et al*.[7] described a locally destructive glomus tumor affecting the nasal cavity under the term 'invasive glomus tumor'. Gaut *et al*.'s case[7] showed erosion of the ethmoid roof to involve the floor of the anterior cranial fossa. One other Korean case, presented by Cho *et al*.[1] also manifested with a large mass (4.5 cm in diameter), but the tumor was described to extend only to the nasopharynx and to be without bony involvement.

Glomangiosarcomas, mostly reported outside the nasal cavity, are composed of round to polygonal tumor cells with single large nucleoli and eosinophilic cytoplasm, forming solid sheets of cells interrupted by vessels.[8] Such features were not observed in our cases.

Our first case closely resembled the Gaut *et al*.'s case,[7] exhibiting the features of cytologically benign glomus tumor but high ki-67 labeling index and invasive feature. Yet we could not use the diagnostic term of 'invasive glomus tumor', since the objective criteria indicating the invasive nature of glomus tumor was almost

Table 1. Sinonasal glomus tumors in the literature

Year	Reference	Age/Sex	Location	Size (cm)	Symptom	Recur
1965	Pantazopoulos[9]	45/F	Inferior turbinate		Obstruction, pain, epistaxis	
1972	DeBord[2]	33/F	Posterior choana		Obstruction	
1974	Fu and Perzin[10]	71/F	Anterior nasal septum		Asymptomatic	
1979	Fleury *et al*.[3]	24/M	Nasal septum		Obstruction	
1984	Potter *et al*.[11]	81/F	Nasal septum		Asymptomatic	
1986	Morais *et al*.[12]	66/M	Nasal vestibulum		Asymptomatic	
1992	Alarcos Llorach *et al*.[6]	55/M	Ethmoid sinus		Obstruction	One recur
1993	Hayes *et al*.[4]	32/F	Nasal vestibulum		Obstruction	Six recur
1997	Arens *et al*.[13]	40/M	Inferior turbinate		Epistaxis	
1998	Shimono *et al*.[14]	55/M	Ethmoid sinus		Obstruction	
1999	Matschiner *et al*.[5]	9/F	Nasal septum	0.5	Epistaxis	
		36/F	Nasal septum		Local pain, bleeding	
		74/F	Nasal septum		Local pain, bleeding, obstruction	
1999	Chu *et al*.[15]	74/F	Nasal region	0.8		
		57/M	Left middle terbinate	0.6		
2000	Nakagawa *et al*.[16]	42/M	Sphenoid sinus	0.4	Obstruction	
2000	Constantinidis *et al*.[17]	66/F	Right middle meatus		Epistaxis	
2000	Cullen and Hanna[18]	50/F	Inferior turbinate	2	Epistaxis	
2003	Ahmed *et al*.[19]	56/F	Nasal septum	0.3	Nasal pain, epistaxis	
2003	Li *et al*.[20]	69/F	Left nasal septum	1	Epistaxis	
2004	Keelawat *et al*.[21]	66/F	Right nasal septum	0.7	Epistaxis	
2005	Gaut *et al*.[7]	87/F	Right posterior choana		Obstruction, epistaxis	
2006	Cho *et al*.[1]	82/M	Nasal septum	4.5	Obstruction, epistaxis	
2008	Current case 1	66/F	Right nasal cavity	3.1	Obstruction, epistaxis	
2008	Current case 2	92/M	Nasal septum		Epistaxis	

F, female; M, male.

lacking.

Other differential diagnoses of nasal glomus tumors are sinonasal hemangiopericytoma, olfactory neuroblastoma, nasal glioma, and embryonal/alveolar rhabdomyosarcoma. However, immunohistochemical demonstration of vimentin and smooth muscle actin is diagnostic of the glomus tumor. The Ki-67 labeling index was high in our first case, reflecting a faster growth than the usual glomus tumors, but its significance is still remained to be elucidated.

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