# CASE REPORT

# Diagnostic challenges of sinonasal extracranial meningiomas: A case report

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Abstract. Background and aim: Meningiomas, the most prevalent non-glial intracranial neoplasms, exhibit different biological features and are typically identified with a characteristic dural tail sign on MRI. However, a subset, approximately 7-16% can manifest extracranially. This case report presents a 70-year-old-woman with nasal dyspnea and intermittent epistaxis due to an ectopic meningioma originating from the nasal cavity, a rare extracranial location. Additionally, a thorough review of the relevant literature was conducted. Methods: Radiological imaging, including maxillofacial CT and MRI, revealed a solid tissue occupying the left nasal cavity, having its origin in middle turbinate. The patient underwent endoscopic sinus surgery (ESS), leading to the histological diagnosis of a WHO grade I meningothelial meningioma with psammomatous bodies. Follow- up at 3-6-12 months showed no recurrence. Results: Extracranial meningiomas constituting less than 1% of non-epithelial tumors, often mimic other sinonasal masses, posing diagnostic challenges. Comprehensive radiological investigation, complete surgical removal, and thorough histopathological examination are crucial for accurate diagnosis and effective treatment planning. Conclusions: This report and literature review emphasizes the importance of preoperative diagnostic-instrumental paths and highlight specific radiological patterns for differential diagnosis, contributing to the understanding and the management of this rare presentation of meningiomas. (www.actabiomedica.it)

**Key words:** sinonasal meningiomas, differential diagnosis, diagnostic pitfalls, skull base tumors, meningioma misdiagnosis, extracranial neoplasms, sinonasal tract tumors, CT/MRI sinonasal meningiomas, histopathology sinonasal meningiomas

### Introduction

Meningioma is the most prevalent non-glial intracranial neoplasm within the central nervous system, categorized into three grades and fifteen histological subtypes by the World Health Organization (WHO) (1). The various types and grades of meningiomas exhibit distinct biological features (2). Typically, meningiomas manifest intracranially, displaying a characteristic dural tail sign on gadolinium-enhanced brain magnetic resonance imaging (MRI). However, occasional cases involve infiltration or extracranial

locations (3). As reported by Rege et al. (4), approximately 7-16% of meningiomas can be found in extracranial sites. Ectopic meningiomas, which display meningioma morphology in tissues and organs without meningeal covering, are categorized into primary and secondary types based on their connection with the central nervous system. Primary heterotopic meningiomas refer to lesions without any association between the tumor tissue and the cranial nerve foramen, vertebral canal, or intracranial structures. These tumors can occur in diverse extracranial locations, such as the middle ear, mouth, nose, parotid gland, neck,

and other regions (5-6). While bilateral obstruction typically suggests nasal polyposis or mucosal inflammation, unilateral obstruction may indicate a structural etiology or a suspected space-occupying lesion. To illustrate, we present a case involving an ectopic meningioma originating from the nasal cavity. This report adheres to the Surgical Case Report (SCARE) Guidelines (7). Additionally, a thorough review of the relevant literature was conducted to provide a comprehensive foundation for the study.

# **Case Report**

A non-smoking 70-year-old woman with no medical history of cranial trauma and head and neck surgical interventions sought ear, nose, and throat (ENT) consultation due to nasal dyspnea that had arisen in the last 12 months, complicated by intermittent epistaxis in the last 45 days. The patient only reported hypertension in her medical history.

#### Ear, Nose, and Throat examination

The endoscopic examination of the upper respiratory tract highlighted a polypoid neoplasm that entirely occupied the left nasal cavity, starting from the left middle turbinate and extending beyond the choana. The lesion appeared to have a pinkish-grayish hue, was not friable, but bled slightly upon palpation. The surface was characterized by a fine vascular network (Figure 1). Inspection and palpation of the nasal pyramid showed no swelling. Additionally, the patient did not exhibit signs of proptosis or visual disturbances.

## Radiological imaging

Firstly, the patient underwent a maxillofacial computed tomography (CT). The CT scan revealed the presence of an expansive lesion occupying the left nasal fossa and contralaterally displacing the nasal septum, resulting in deformity and thinning of the medial wall of the left maxillary sinus, which appeared to be interrupted at several points (Figure 2). Also, the lesion extended into the nasopharynx and was characterized by internal calcifications but did not show



**Figure 1.** This endoscopic image reveals a pink-grey mass within the left nasal cavity, emerging from the middle turbinate and fully occupying the nasal fossa, resulting in near-complete obstruction. The lesion's surface exhibits a smooth texture characterized by a superficial capillary network.

infiltrative-type radiological phenomena, while the turbinates appeared difficult to assess. The diameter of the neoplasm was 4.6 cm antero-posteriorly and 3.5 cm caudally with nasal septum dislocation, bowing of the medial wall of the maxillary sinus, associated bone erosion, nasopharynx extension, and no infiltration pattern. The ipsilateral maxillary and sphenoid sinus were occupied by isodense tissue. The maxillofacial MRI examination with contrast (Figure 3) showed signal alteration corresponding to solid tissue that occupied a large part of the left nasal cavity with probable origin from the middle turbinate. The alteration appeared isointense in T1 and hyperintense in T2 showing significant enhancement in T1 after administration of contrast agent. In the DWI sequences, the lesion appeared isointense, slightly hyperintense on FLAIR, with elevated values on ADC.

## Surgical treatment

Before surgery, the diagnostic hypotheses were degenerated antrochoanal polyp (unlikely), inverted



**Figure 2.** This CT scan illustrates the lesion encompassing the entire left nasal fossa and the corresponding maxillary sinus. The middle turbinate is indiscernible, the medial wall of the maxillary sinus shows signs of erosion, and the nasal septum is deviated to the contralateral side. Additionally, scattered microcalcifications are evident.

papilloma, carcinoma, or juvenile fibroangioma (though epidemiologically less likely) or meningoencephalocele. For this reason, despite the preoperative biopsy potentially being necessary to establish a correct therapeutic pathway, the risk of vascular lesions or encephalic herniation led to the decision for a primary surgical approach. The patient underwent endoscopic sinus surgery (ESS) for the removal of the neoplasm arising from the left middle turbinate, which was partially sacrificed. Uncinectomy and antrostomy were performed concurrently with drainage of purulent exudate from the ipsilateral maxillary sinus. Subsequent sending of biopsy sample for definitive histological examination.

## Histology

Histopathological examination revealed an unencapsulated tumor composed of tumor cells arranged in nests with whorling patterns and sparse psammoma

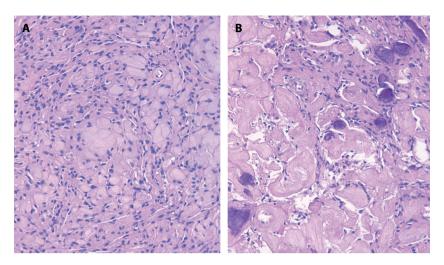


**Figure 3.** Contrast-enhanced maxillofacial MRI displaying signal changes suggestive of solid tissue within a significant portion of the left nasal cavity. The origin is likely the middle turbinate, as observed to be isointense in T1 and hyperintense in T2 accompanied by irregular pathological contrast impregnation.

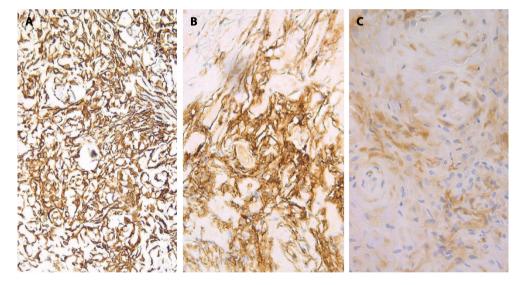
bodies (Figure 4). The tumor cells were epithelioid to spindle-shaped with monomorphic round nuclei, a moderate amount of eosinophilic cytoplasm, and indistinct cell borders. There was an absence of nuclear atypia, significant mitotic activity, and necrosis. Immunohistochemistry showed positivity for Vimentin, Epithelial Membrane Antigen (EMA), and S-100 protein (Figure 5) and the absence of a reaction for cytokeratins, CD34, and smooth muscle actin. Based on these findings, a diagnosis of a WHO grade I meningothelial meningioma with psammomatous bodies was given. The differential diagnosis considered and excluded angiofibroma, cement ossifying fibroma, paraganglioma, and schwannoma.

# Follow-up

No recurrence was found at the post-op followup at 3, 6, and 12 months.



**Figure 4.** Whorled growth pattern of meningothelial spindle cells (A) accompanied by psammoma bodies (B) (Hematoxylin and Eosin stain, magnification X200).



**Figure 5.** Immunohistochemistry showing positive stains for Vimentin (A), EMA (B), and S-100 protein (C).

#### Discussion

Our clinical case begins with progressively worsening dyspnea for approximately a year, associated with recurrent epistaxis that started a month ago. Furthermore, on the CT scan, the lesion occupies the nasal fossa and deviates the nasal septum, with attachment at the level of the middle turbinate. The radiological pattern rules out the presence of bone infiltration but reveals intralesional calcifications. Furthermore, the most

associated histological pattern with sinonasal meningiomas is the meningothelial type. In our case, there is also an association with the meningothelial histological type and the presence of psammomatous bodies. The studies conducted on clinical cases like those addressed in our center highlight the importance of identifying an appropriate preoperative diagnostic-instrumental path. This is essential for conducting a correct staging of the lesion and ensuring a more effective treatment. Particularly, the purpose of this report is to focus on

the patient's physiological and surgical history, clinical evaluation, and endoscopic appearance of the lesions. Additionally, we will seek to clarify the role of radiological diagnostics and the guiding symptoms that lead to a correct diagnosis. Meningiomas are nonglial tumors of the central nervous system, representing 24–30% of all intracranial neoplasms. They have been reported to occur extracranially in only 1-2% of cases, and 20% of extracranial meningiomas are secondary extensions of intracranial tumors. Primary extracranial meningiomas without direct communication with the intracranial region are rare. Histologically, primary extracranial meningiomas are identical to intracranial counterparts (8-9).

An analysis of 146 cases of primary extracranial meningiomas showed that most of them originated from the skin and scalp (n = 59) followed by the middle ear (n = 26) and sinonasal tract (n = 25) (10).

Patients with sinonasal localization exhibited nasal obstruction and congestion, rhinorrhea, headache, and a sense of pressure. Sometimes, intermittent epistaxis and progressive obstruction can be observed, leading to a gradual worsening of symptoms (11). The propensity of meningiomas to infiltrate the bone and spread through the Haversian canals, along with spread through natural foramina in the skull base, allows for the involvement of both intracranial and extracranial sites (12). Significant hyperostosis has been recorded, which can extend up to the nasal septum. Zhang et al. (13) reported four cases of hyperostosis and three cases of bone destruction in a series of 43 patients having anterior cranial fossa meningiomas invading PNS and NC. In Table 1, we have indeed compared the most recent and numerous studies in the literature, analyzing which aspects could be fundamental for a correct differential diagnosis such as demographics, clinical presentation, radiological imaging, histology, performed treatment, and the definitive diagnosis. In addition to what has been emphasized regarding the clinical presentation and the site of origin of the lesion, we believe that a correct identification of specific radiological patterns could hold significant value (14-21).

From a clinical standpoint, unilateral nasal obstruction and episodic epistaxis emerge as the most common symptoms (14,16-19,21). Additionally, the lesion often appears grayish, completely occupying one nasal cavity with an imprint on the nasal septum

and contralateral displacement (14,18,19,21). The most frequently consulted radiological image is the contrast-enhanced CT scan, which often highlights bone erosion and concurrent intralesional microcalcifications (14-21). Angiography was performed in only one case, suspected of juvenile fibroangioma (15). MRI more frequently reveals a lesion that is isointense on T1-weighted images and heterogeneously hyperintense on T2-weighted images (15,17,18,20).

The preferred surgical approach has been the endoscopic one, with open surgery chosen in only two cases. In one case, (18) a preoperative diagnostic biopsy was performed, and in another case (19), arterial embolization of the internal maxillary artery was carried out before endoscopic surgery. The definitive diagnosis has consistently been made based on histological examination, particularly immunohistochemistry, which reveals frequent positivity for Vimentin, EMA (Epithelial Membrane Antigen), and S-100 (14-21). Grade I meningothelial meningioma according to the WHO classification was most reported. The distinctiveness of our case stems from the unusual origin of the lesion, which is atypically situated on the middle turbinate. Notably, the patient had no prior history of surgery, and the sole discernible and persistent symptom was headaches, particularly associated with episodes of epistaxis. The identification of psammomatous bodies in the final histological examination is a seldom-reported occurrence. After the surgical intervention, serial endoscopic examinations were performed every four months for follow-up. At the last check-up, approximately one year after the surgery, the nasal cavity appeared patent, with well-stabilized results from the previous surgery and no presence of exudate or locoregional signs of recurrence (Figure 6-7).

## **Conclusions**

Meningiomas are the most common neoplasms of the central nervous system, constituting approximately 25% of intracranial tumors. Although they are typically identified in older individuals, they can occur at any age and are more prevalent in women. Ectopic meningiomas, accounting for less than 1% of non-epithelial tumors, are rare (22). Primary extracranial meningiomas are sporadically reported and primarily involve

Table 1. The table presents case reports from the literature stratified by age, gender, symptoms, radiological exams, surgical treatment, histology, and final diagnosis.

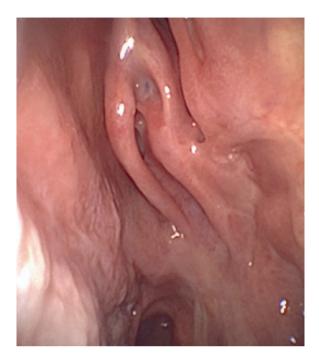
	,		,			ENT			,		
	Authors	Year	Age	Sex	Symptoms	examination	CT	MRI	Histology	Treatment	Diagnosis
1	J.K. Sharma <sup>14</sup>	2006	13	M	Unilateral nasal	Swelling of the	Homogeneously		Spindle cells	Lateral	Psammomatous
,					obstruction	side face	enhancing mass			rhinotomy	meningioma
					Episodic	Pinkish	Right ethmoid		Oval blend	Moore's	
					epistaxis	polypoidal mass	sinus Nasal cavity		nuclei	incision	
					Proptosis	filling the right	lateral		Many	Nasal	
					4	nasal cavity	displacement of		psammoma	septum was	
						•	the medial wall		bodies	repositioned	
						- -	OI rigiit orbit			Dack	
						Pushing the	Compression of		Multifold		
						septum to the left	right maxillary		calcification		
						peeping	Deviation of				
						through the	nasal septum				
						choana					
2	K. Kainuma <sup>15</sup>	2007	42	M	Headache	Originating from ethmoid sinuses	Mass extending from the right ethmoid sinuses	Enhanced mass	EMA positive	Endonasal approach	Meningothelial meningioma (grade I)
											/
					Temporary visual disorder	Involving: nasal cavity and sphenoid sinus	Bone erosion through the posterior skull	Angiography: moderate feeding arteries (from		Navigation system	ОНМ
							base	CE)			
3	M. Mnejja <sup>16</sup>	2011	18	ഥ	Unilateral nasal obstruction	Grayish, firm, nonbleeding	Soft tissue dense mass		EMA positive	Endonasal approach	Meningothelial meningioma
						mass	obstructing nasal cavity				(grade I)
					Slowly growing	Origin: middle	filling of the		Vimentine		WHO
					0	meatus	homolateral		positive		
							anterior				
							ethmoidal cells				
							and maxillary				
							Laminated		S100, Keratin,		
							orbit		CEA focal		
									immunoreactivity		

	Authore	Voor	Ασο	Š	Summtome	ENT	CT	MBI	Histology	Treatment	Diamoeie
	A WELLOLD	ICAI	787	Y .	cymproms	Commination		TATIO	ristorogy	, , , , , ,	L'agnosis
4	D. Mondal	2015	7	Z	Episodic	Swelling of the	Hyperdense	Isointense	Vimentine,	Endonasal	Meningothelial
					epistaxis	nasal bridge	mass in the	to mildly	S100, Ki67	approach	meningioma
							ethmoid sinus	hypointense on	positive		(grade I)
					·	-	מוות וומסמו כמיווי)				OI III
					Left eye watering	Fleshy, firm, bleeding to	homogeneous enhancement of	Iso- to hyper intense on T1-W			МНО
					S	touch mass	the mass				
					Proptosis, optic	Developed from	Bony erosion				
				]	neuropathy,	ethmoid sinus	and expansion				
					restricted ocular		No intracranial				
					movement		component				
Ŋ	R. Wadhera <sup>18</sup>	2016	55	ഥ	Unilateral nasal	Nasal mass in	Soft tissue	Mass extended	Meningo-	Nasal biopsy	Primary naso-
							sinus	ethmoid sinuses		under local anesthesia	meningioma
					Episodic	Filling nasal	Involving	Iso-intense	Psammoma	Lateral	WHO
					epistaxis	cavity	septum and medial wall of left orbit	M1	bodies	rhinotomy incision	
						Dushing		Heterogeneously	Nuclear	Mass	
						t usumig the septum		high signal on	membrane	excision	
						contralaterally		T2W	progesterone	(superior	
								Short TI Inversion	receptor positive	attachment	
								Recovery (STIR)	positive	,Smm	
								images.	S-100, Keratin,		
									CEA tocally positive		
9	L. Maharjan <sup>19</sup>	2018	63	ഥ	Unilateral nasal	Pinkish fleshy	Nasal cavity		Tumor cells	Preoperative	Atypical
					obstruction	mass	heterogeneously		arranged in	internal	transitional
							enhancing mass		lobules, nests, and sheets	maxillary	meningioma (grade II)
					Episodic	Filling nasal	Extension into		monomorphic	Artery	WHO
					epistaxis	cavity	ethmoid sinus,		round nuclei	embolization	
							choana and				
							sphenoid				
							Erosion of the		Mitotic figure:	Endonasal	
							ardanciii coiic			The Land	
							Destruction of		S-100 positive		
							me mom ochemi				

	Yea	H	Age	Sex	Symptoms	ENT examination	CT	MRI	Histology	Treatment	Diagnosis
L.C. Fechtner 2021 64		64		Ŀı	Intellectual disability, seizures	Left nasal dorsum palpable mass. Proptosis and telecanthus.	IV contrast: homogenously enhancing lobulated mass.	Hypointense on T1	Low-grade neoplasm	Weber- Ferguson incision as well as bicoronal flap	Atypical meningioma with bone invasion
							Extension: midline anterior cranial fossa	Mildly hyperintense on T2	EMA weakly positive	Repair with pericranium and fascia lata grafts	
							Including olfactory groove, frontal sinus, and anterior ethmoid sinus	Displaced homogeneous avid enhancement	S100 diffusely positive		
							Extending into bilateral middle meatus and left anterior nasal Cavity		CK7, CD99 positive	NO RT	
							marked scalloping Thinning of the bone				
M. Abir <sup>21</sup> 2022 41		41		M	Unilateral nasal obstruction	Well circumscribed soft tissue mass	Nasal cavity homogeneously enhancing mass		Spindly cells with oval bland nuclei	Endonasal approach	Meningothelial meningioma (grade I)
		_			Facial pain	filling nasal cavity	Extension: ethmoid sinus superiorly		Moderate amount of eosinophilic cytoplasm		МНО
					Episodic epistaxis	Pushing the septum contralaterally	erosion of adjacent bone		Vimentin, S 100, Ki67 positive		

Meningothelial meningioma (grade I)	(with psammomatous bodies)	WHO		
Endonasal approach (ESS)				
Spindle- shaped with monomorphic round nuclei	Vimentin, EMA, S-100 positive			
Isointense in T1	Hyperintense in T2	Irregular pathological contrast impregnation.		
Expansive lesion occupying the left nasal fossa	Contralaterally Hyperintense displacing the in T2 nasal septum	thinning of the medial wall of the left maxillary sinus	Extension into the nasopharynx	Internal calcifications
Filling nasal cavity	Origin: left middle turbinate	peeping through the choana		
Nasal dyspnea	Epistaxis			
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Abbreviations: M (male), F (female), EMA (Epithelial membrane antigen), WHO (World Health Organization), CE (contrast enhancement), CEA (carcinoembryonic antigen,), HPF (high power field), CK7 (Cytokeratin 7), RT (radiotherapy).



**Figure 6.** Endoscopic image of the left nasal cavity patency. The remnant of the middle turbinate is visible.



**Figure 7.** A detailed view of the remnant of the middle turbinate and the axilla is visible.

the cranial and cervical regions (23,24). Meningiomas are frequently misdiagnosed as nasal polyps or inverted papillomas and this can make diagnosis challenging (25). Radiological imaging is crucial for preoperative

planning. A combination of CT, MRI, and angiography aids in assessing the tumor's extension and involvement of major vascular structures (26). The definitive diagnosis of extracranial meningioma is made through histopathological analysis and surgery is the primary treatment for extracranial meningiomas, with radiotherapy considered in selected cases or for recurrence (27). In conclusion, diagnosing adult cases of primary nasal meningiomas can be challenging due to their mimicry of other sinonasal masses. Comprehensive radiological investigations, complete surgical removal, and thorough histopathological examinations are essential for accurate diagnosis and effective treatment planning.

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Note: The informed consent was obtained for the surgical procedure. In compliance with the relevant legislation, data processing in this study was conducted under the provisions of Article 110-bis of the Italian Privacy Code (Legislative Decree No. 196 of June 30, 2003). The legislation can be accessed at (https://www.brocardi.it/codice-della-privacy/parte-ii/titolo-vii/capo-iii/art110bis.html).

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