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**LETTER TO THE EDITOR**

# Osteoma infiltrated by schwannoma in sinonasal region: radiologically masquerading as a malign tumor

## Sinonasal bölgede schvannom tarafından infiltre edilen osteom: radyolojik malign kuşkulu tümör

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To the Editor;

Tumours of the sinonasal cavity and paranasal sinuses mostly develop from mucosal epithelium, seromucous glands, soft tissues, bone, cartilage, neural/neuroectodermal tissue, hematolymphoid cells and the odontogenic tissues. However neoplasms of sinonasal cavity are dominated by epithelial type[1]. Benign epithelial tumors frequently seen in sinonasal cavity include papillomas and salivary gland-type adenomas. Benign soft-tissue neoplasms of the sinonasal cavity are uncommon and include nerve-sheath tumors, hemangioma, angiofibroma, myxoma, leiomyoma, and meningioma. Here, we presented co-occurence of osteoma and schwannoma. So far, osteoma infiltrated by schwannoma in sinonasal region is not reported in English literature. Additionally it was masquerading as malignant tumor radiologically.

A 63 years old man complained of progressive nasal obstruction and headache. Computed tomography (CT) scan showed a mass lesion in size of 56x21 mm that was filling the left ethmoid cells, nasal cavity and sphenoid sinus. The lesion had osseous component in size of 40x27 mm at the level of ethmoid cells and solid components at level of inferior nasal cavity (Figure 1G,H,J). Radiologic diagnosis was osteosarcoma. After complete surgical excision, during five years of follow-up, there was no recurrence. Microscopic examination revealed a dense mature bone that was sharply demarcated from mucosal respiratory epithelium (Figure 1C). The interosseous and peripheral stroma was loosely arranged and edematous. There was also an accompanying spindle cell neoplasm with vaguely hypocellular and hypercellular arrangement in interosseous and peripheral stroma (Figure 1 A-B and D). Spindle cells showed no atypia, typical verocay bodies, necrosis, and mitosis. Immunohistochemical studies showed that spindle cells were positive for S-100 (Figure 1E-F) and vimentin.

Schwannoma is a benign soft-tissue tumor evolving from the schwann cell of the neural sheath, and 25 to 45% occur in the head and neck[2]. Less than 4% of head and neck schwannoma occurs in the nasal cavity and paranasal sinuses[3]. Schwannoma of sinonasal tract originates from branches of the trigeminal nerve[4]. The ethmoid sinus is most commonly involved. Maxillary sinus, nasal fossa and sphenoid sinus are less commonly involved[5]. There are no predilection age, race, or gender in sinonasal schwannomas. Most common symptoms are nasal obstruction, nasal discharge, and anosmia. Sinonasal osteoma was first reported by Viega[6]. Osteoma is reported to be the most common benign tumor of sinonasal tract. It is most frequently located in the frontal sinus (%57), followed by the maxillary sinus, ethmoid and sphenoid sinuses[6]. However, it is rare in the nasal cavity. Osteoma is usually asymptomatic given their slow growing nature. Sinonasal osteomas occur with clinical findings related to mass effect

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such as headache and facial distortion. Unlike schwannomas in other localizations, sinonasal schwannoma is distinguished by the absence of a peripheral capsule[7]. Sinonasal schwannoma's clinical symptomas are varied and nonspecific, frequently in relation with signs of chronic nasal obstruction.

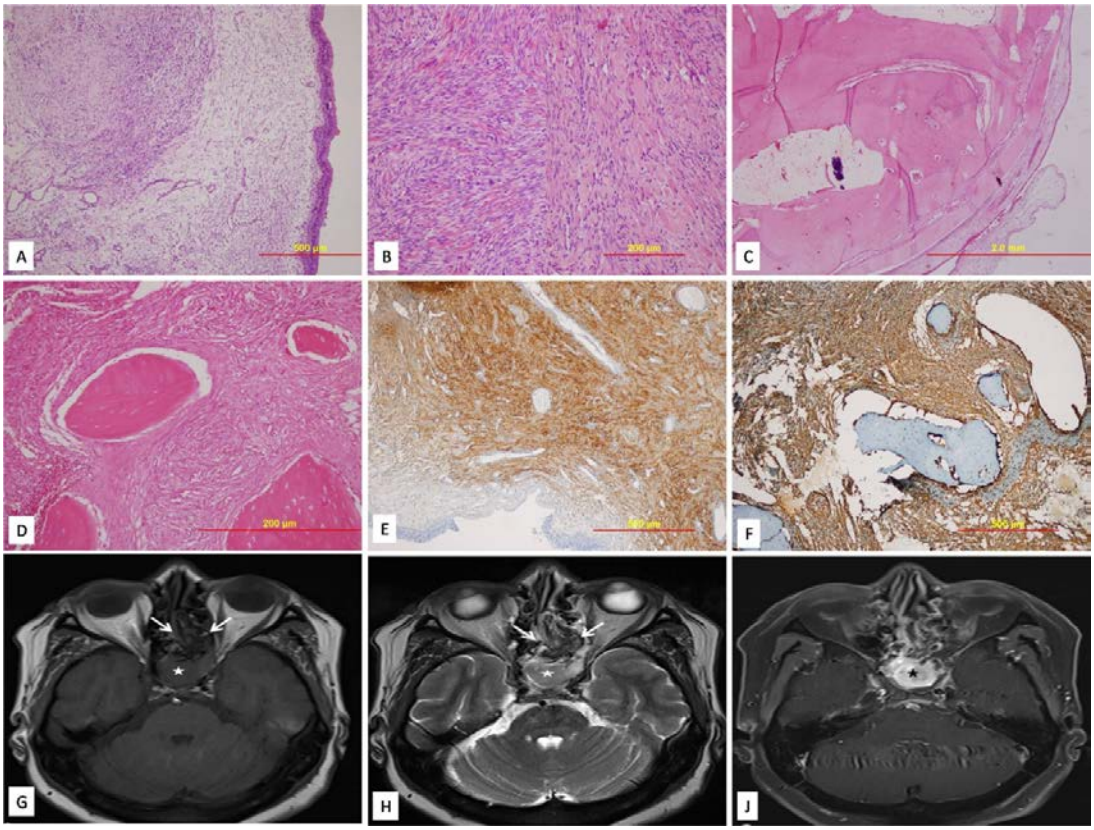


**Figure 1. A (HEX100) Spindle cell neoplasm with vaguely hypocellular and hypercellular arrangement under the mucosal respiratory epithelium. B (HEX200) Some areas are cellular (left side), some areas are fibrotic (right side).C (HEX40) Dense mature bone formed neoplasm that was sharply demarcated from mucosal respiratory epithelium. D (HEX200) Spindle cells are in interosseous areas. E,F (S-100X100) Spindle cells for S-100 positive in peripheral and interosseous stroma. G,H,J (CT figures) Axial pre-contrast T1 and T2-weighted images (G,H) show a mass with a solid component isointense to the cortex of the brain (white stars) and a hypointense osseous component (arrows). Post-contrast axial T1-weighted images (J) show enhancing solid component and extension of the mass to anterior cranial fossa (black stars).**

In our case, the first biopsy showed that the lesion was composed of mature bone tissue. It was reported to be compatible with osteoma. After total surgical resection, it was seen that the lesion had also a mesenchymal component consisting of spindle cells. It was seen that this mesenchymal component infiltrated between the mature bones trabecule of the osteoma and also progressing under the mucosa. The lesion was unencapsulated with ill-defined borders. Hyalinized vessels or verocay bodies were not seen.

Osteomas are generally detected incidentally during radiological evaluation. The pathologist should be careful when radiological diagnosis is malignant in such benign bone lesions. Hypocellular edematous mesenchymal tissue, which may be accompanied to benign bone lesion may represent a possible hypocellular areas of schwannoma. In this regard, it may be useful to evaluate S-100 protein to rule out schwannoma. On the other hand, the spindle cell areas associated with osteoma may be suspicious for malignancy. For this reason, cellular atypia, necrosis,

mitotic activity, infiltration of the sinonasal submucosa, and osseous invasion should be carefully evaluated. Schwannoma of the sinonasal cavity should be differentiated from malignant peripheral nerve sheath neoplasm[8]. It also should be differentiated from the osteosarcoma. Therefore, the new bone formation produced by atypical cells is needed for diagnosis of osteosarcoma.

Meningioma, angiofibroma, neurofibroma, and leiomyoma should be considered in differential diagnosis of schwannoma[9]. Neurofibroma is composed of schwann cells, perineural cells and intraneural fibroblasts. Immunohistochemically neurofibromas show low reactivity to S-100 protein than schwannomas. It can also show reactivity for EMA and CD34. Meningioma can also seen in sinonasal cavity. Meningioma cells shows syncytial setting, whorled pattern and include pseudonuclear inclusions. Meningioma can also has psammoma bodies. Positivity for EMA can be helpful to distinguish of meningioma. Angiofibroma have hyalinized vascular stroma containing stellate and spindled myofibroblasts. There are also numerous mast cells in angiofibroma. The vascular structures have variable wall thickness and characterized by staghorn appearance. Leiomyoma grows in intersecting fascicles of spindled cells. Immunohistochemically it reacts for desmin, SMA, calponin and H-caldesmon.

In conclusion benign bone lesions such as osteoma are common in the sinonasal region, whereas schwannomas are rare. Schwannomas of the sinonasal region have irreguler borders with no capsule. In this region, the biopsy samples taken from radiologically suspected bone lesions may be accompanied by areas of hypocellular or hypercellular spindle cells. These cases with spinal cell areas should be evaluated with immunohistochemically for S-100 proetein for the possibility of Schwannoma. For this reason, the pathologist should be aware of a second soft-tissue tumor that may accompany the radiologically suspicious malignant lesions. We found it worth to report the coexistence of Schwannoma with osteoma which may be radiologically suspicious for malignancy and can be overlooked in microscopic examination.

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