CASE REPORT

https://doi.org/10.5125/jkaoms.2018.44.1.29
pISSN 2234-7550 · eISSN 2234-5930

Primary sinonasal mucosal melanoma simulated as cystic lesions: a case report

Sung-Ho Shin[1], Hyun Seok[1,2], Seong-Gon Kim[1], Seong-Doo Hong[3]

[1]*Department of Oral and Maxillofacial Surgery, College of Dentistry, Gangneung-Wonju National University, Gangneung,*
[2]*Department of Oral and Maxillofacial Surgery, Chungbuk National University Hospital, Cheongju,*
[3]*Department of Oral Pathology, School of Dentistry, Seoul National University, Seoul, Korea*

Abstract (J Korean Assoc Oral Maxillofac Surg 2018;44:29-33)

Sinonasal mucosal melanoma (SNMM) in the maxillary sinus is a rare disease condition. Compared to oral mucosal melanoma, SNMM has a bulky, exophytic, and polypoid appearance, is weakly pigmented, and associated with unspecific symptoms. Due to these features, SNMM in the maxillary sinus has been misdiagnosed as nasal polyps and chronic sinusitis. In this case report, we described SNMM occurring in the right maxillary sinus simulated as a cystic or benign lesion. Cortical bone thinning and expansion were observed around the mass. The excised soft mass was encapsulated and weakly pigmented. The mass was clearly excised and covered with a pedicled buccal fat pad graft. Diagnosis using immunohistochemistry with S-100 and homatropine methylbromide-45 (HMB-45) is critical for proper treatment.

Key words: Sinonasal mucosal melanoma, Maxillary sinus, Buccal fat pad, S-100, Homatropine methylbromide-45

[paper submitted 2017. 6. 14 / revised 2017. 7. 19 / accepted 2017. 8. 16]

I. Introduction

Malignant melanomas are classified as mucosal and cutaneous melanomas[1]. Mucosal malignant melanoma is a relatively rare disease compared to cutaneous melanoma that affects the head and neck region[2]. Mucosal malignant melanoma accounts for 0.4% to 2% of all malignant melanomas and 10% of all head and neck melanomas[3]. Involved sites of mucosal melanoma include the oral cavity, nasal cavity, paranasal sinus, nasopharynx, and larynx[4,5]. Sinonasal mucosal melanoma (SNMM) accounts for 3.5% of all malignancy neoplasms found in the sinonasal region and accounts for 6.7% of head and neck melanomas[6]. Melanoma arising from mucosa tends to be more aggressive and has a poorer prognosis than cutaneous melanoma[7]. The 5-year survival rates of SNMM are reported to vary from 0% to 30%[8].

The main site of SNMM is the nasal cavity including the nasal septum and lateral nasal wall[9]. Cases involving paranasal sinuses such as the maxillary sinus and ethmoid sinus are less common[10]. Symptoms of SNMM include nasal blockages, facial pressure, and nasal blood discharge[11]. The etiology of SNMM is unclear[10]. Oral mucosa melanoma has a flat and thin appearance while SNMM manifests as an exophytic, bulky, and polypoid mass[4]. More than one third of SNMMs are characterized by weak or no pigmentation[4,8]. Consequently, diagnosis is difficult and often confused with other pathologies such as nasal polyposis and sinusitis[9]. For an accurate diagnosis, excisional biopsies and histopathological examinations should be required[4]. Immunohistochemical stains with S-100 and homatropine methylbromide-45 (HBM-45) have been performed to confirm diagnosis of SNMM in the head and neck region[12]. We report a case of SNMM primarily occurring in a right maxillary sinus mucosa simulated as a cystic lesion. This case report was approved by the Institutional Review Board (IRB) of Gangneung-Wonju National University Dental Hospital, Gangneung, Korea (IRB 2017-002).

Hyun Seok
Department of Oral and Maxillofacial Surgery, Chungbuk National University Hospital, 776 Isunhwan-ro, Seowon-gu, Cheongju 28644, Korea
TEL: +82-43-269-3139 FAX: +82-43-269-6387
E-mail: sok8585@hanmail.net
ORCID: http://orcid.org/0000-0002-5278-3807

II. Case Report

A 78-year-old female patient was referred from a local clinic for a cystic lesion on her right maxillary sinus. She had facial swelling on the right paranasal and maxillary molar area that formed 2 months prior. Swelling and fluctuation were observed in the right maxilla buccal mucosa without pigmentation.(Fig. 1. A) She suffered from pain and a fever and had no specific medical history except for hypertension. Haziness in the right maxillary sinus was observed on panoramic view.(Fig. 1. B) The large soft tissue mass was also observed via cone-beam computed tomography.(Fig. 1. C) Cortical thinning and expansion of the sinus wall was observed. The medial wall of the sinus had deviated to the nasal cavity. Although the perforation of the cortical bone was observed on the anterior maxillary sinus wall, there was no destructive bony formation or invasive extension to the surrounding structure.(Fig. 1. D)

The soft tissue mass was excised under general anesthesia. The mass extended from the floor of the maxillary sinus to the ostium of the maxillary sinus and medially to the nasal cavity. The tumor mass was well encapsulated within the epithelium and did not infiltrate the medial wall of the maxillary sinus.(Fig. 2. A) The mass was easily separated from the expanded bony wall and was excised in 1 piece. The excised mass was greyish and brownish in color and weakly pigmented. The bony defect was covered with 2 layers of flaps that included the buccal mucosa and pedicled buccal fat pad (BFP).(Fig. 2. B) The specimen included a greyish and brownish soft tissue mass 5.5×4.0 cm in size that was weakly

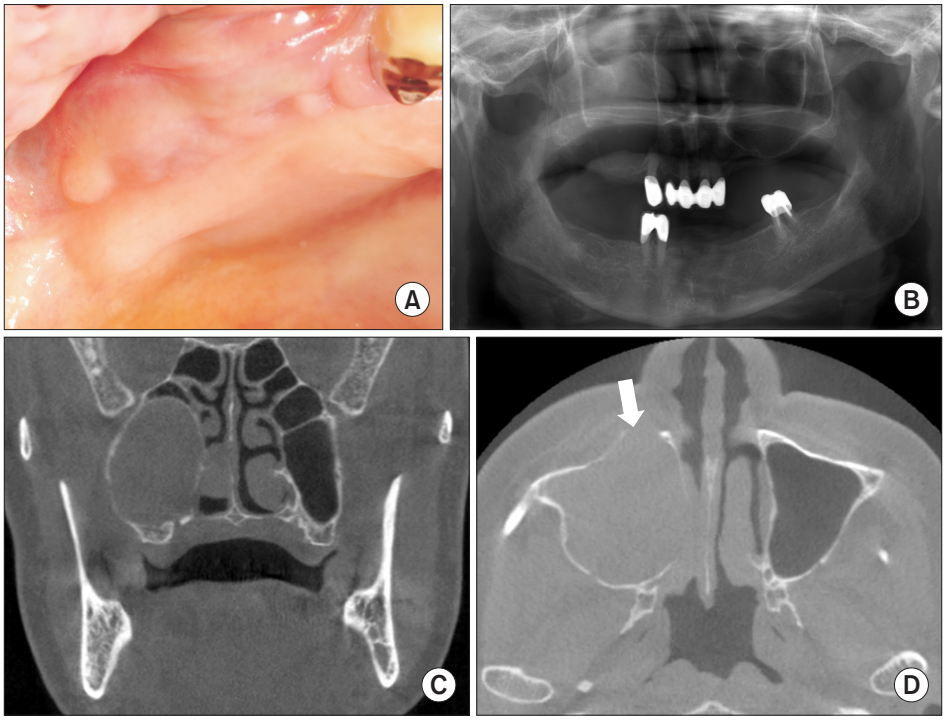

Fig. 1. Radiological and clinical images of the right maxillary sinus. A. Buccal mucosal swelling without pigmentation as result of tumor expansion. B. Haziness of the right maxillary sinus and an indistinct alveolar crestal line on panoramic view. C. Cortical bone thinning and expansion on the right maxillary sinus. D. Cortical bone perforation on the anterior wall of the maxillary sinus (white arrow).
Sung-Ho Shin et al: Primary sinonasal mucosal melanoma simulated as cystic lesions: a case report. J Korean Assoc Oral Maxillofac Surg 2018

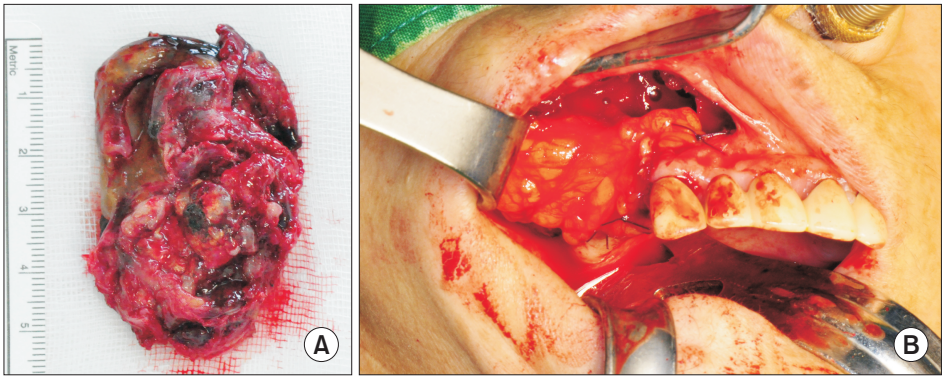

Fig. 2. A. Well encapsulated and partly pigmented soft tissue mass. B. Buccal fat pad graft after tumor excision.
Sung-Ho Shin et al: Primary sinonasal mucosal melanoma simulated as cystic lesions: a case report. J Korean Assoc Oral Maxillofac Surg 2018

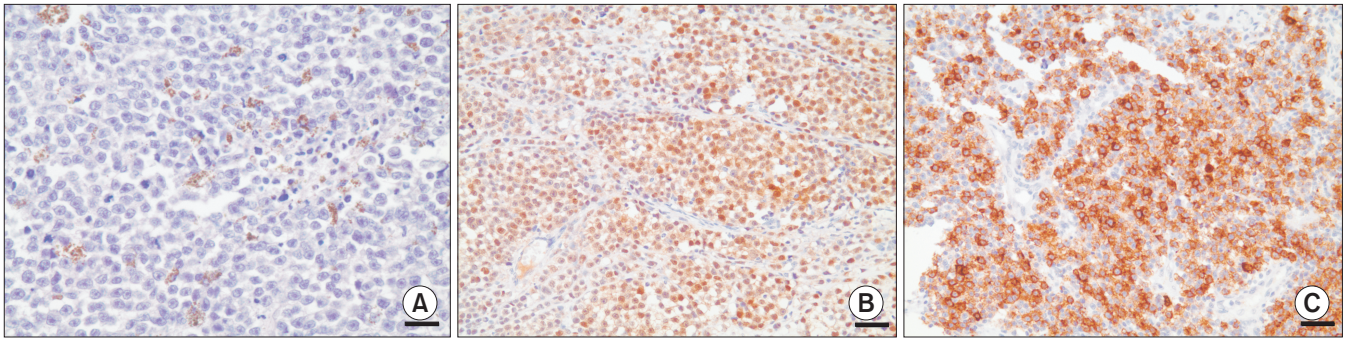

Fig. 3. Histological and immunohistochemical image. Melanin pigmentation and mitosis is observed (A; H&E staining, ×400, scale bar=25 μm). Immunohistochemistry with S-100 (B; ×200, scale bar=50 μm) and homatropine methylbromide-45 (C; ×200, scale bar=50 μm).
Sung-Ho Shin et al: Primary sinonasal mucosal melanoma simulated as cystic lesions: a case report. J Korean Assoc Oral Maxillofac Surg 2018

pigmented.(Fig. 2. A) The tissue mass contained an organized blood clot and hemorrhage.(Fig. 2. A) On histopathological examination, neoplastic tumor cells with prominent nucleoli and malignant spindle cells were observed. Some tumor cells contained melanin.(Fig. 3. A) Based on strongly positive immunohistochemistry results using S-100 and HBM-45, malignant mucosal melanoma was diagnosed.(Fig. 3. B, 3. C) One month after the operation, the surgical site was completely healed without complications and we referred the patient for chemotherapy and radiotherapy treatment. However, she refused all additional treatments and only received some antibiotics.

III. Discussion

SNMMs are malignant neoplasms found in the head and neck region[6]. They typically form in the nasal septum and lateral nasal wall and later in the inferior turbinate, maxillary sinus, and ethmoid sinus[1,10]. SNMMs developing from the paranasal sinus are rarely found[8]. SNMMs have an ulcerative and necrotized surface of variable coloring depending on the characteristics of melanin pigmentation[10]. They cause unspecific symptoms such as nasal obstructions, recurrent nasal epistaxis, and sometimes facial pressure[11]. Recently, such symptoms have been detected in the maxillary sinus[11]. Due to the nonspecific and late onset of these symptoms, diagnosis of maxillary sinus SNMM is difficult[1]. In our case, the patient did not experience nasal epistaxis or obstruction. She exhibited facial swelling and fluctuations in the right maxillary buccal mucosa due to the mass.

SNMMs in the maxillary sinus grow aggressively and invasively[11]. They extend invasively and infiltrate surrounding structures such as the nasal cavity, ethmoid sinus, and middle turbinate[1]. Radiologically, SNMMs have an ill-defined border and show signs of bone destruction around a lesion[11]. However, low-grade SNMMs can manifest as a polypoid mass without infiltrating lesions and are often confused with nasal polyps, chronic sinusitis, and inflammatory pseudotumors[9,13-15]. In our case, a large soft tissue mass was observed on the right maxillary sinus with round appearance. Cortical bone thinning and mass expansion to the nasal cavity were observed.(Fig. 1. C) Although a cortical bone perforation was observed on the anterior sinus wall, signs of bone destruction or irregular margins around the mass were not present.(Fig. 1. D) Consequently, this case was difficult to identify and initially diagnosed as a cystic or benign lesion.

SNMMs are difficult to diagnose due to the presence of variable neoplastic cells[2]. SNMMs have small blue round cells, pleomorphic and epithelioid cells, and spindle cells[10,16]. Due to the presence of round blue cells, SNMMs can be confused with lymphomas, olfactory neuroblastomas, or small cell carcinomas[10]. Pleomorphic and epithelioid cells may be misdiagnosed as carcinoma. Melanoma spindle cells are also often confused as sarcomas or spindle cell carcinomas[9]. Malignant melanoma is diagnosed based on the presence of melanin pigmentation in tumor cells[1]. However, more than one third of SNMMs lack pigmentation and are referred to as amelanotic melanomas[14]. For these reasons, immunohistochemical staining is necessary for the diagnosis of SNMM.

Expressions of S-100 and HBM-45 were used as diagnostic markers for melanoma[17]. S-100 protein is derived from Schwann cells, melanocytes, and myoepithelial cells. The protein is found in cases of melanoma, schwannoma, and neurofibroma[16]. HMB-45 is a monoclonal antibody that reacts with the antigen derived from melanoma[16]. Histologically, neoplastic tumor cells were mostly dispersed and grew with prominent nucleoli. Furthermore, melanin pigmentation was observed in the cytoplasm of tumor cells.(Fig. 3. A) Based on

immunohistochemistry analysis, the specimens were positive for S-100 and HBM45.(Fig. 3. B, 3. C) Consequently, the patient was diagnosed with primary SNMM from the maxillary sinus mucosa.

SNMMs have a poorer survival rate when found in the maxillary and ethmoid sinuses than in the nasal cavity[11]. SNMMs in the maxillary sinus are closer to the skull base and can easily infiltrate the lamina papyracea[11]. A malignancy of the skull base and orbit is associated with poor prognosis[18]. Fortunately, our case did not involve the skull base but instead expansion to the ostium and anterior wall of the maxillary sinus.(Fig. 1. C) Standard SNMM treatment involves broad surgical excision with negative margins[10]. Depending on the size of the tumor and the degree of lymphatic invasion additional chemotherapy and radiation therapy may be required[1,19]. In our case, the tumor was well encapsulated and thus was easily excised; the defect was covered with pedicle BFP to prevent fistula formation.(Fig. 2. B) BFP grafting is a useful method for covering intraoral defects after tumor resection[20].

In our case, a primary SNMM was found arising from the maxillary sinus mucosa simulating a cystic lesion. We diagnosed the SNMM based on the histological features of melanin pigmentation in the neoplastic tumor cells. The final diagnosis was established via immunohistochemistry with S-100 and HMB-45.

ORCID

Sung-Ho Shin, *http://orcid.org/0000-0001-6995-373X*
Hyun Seok, *http://orcid.org/0000-0002-5278-3807*
Seong-Gon Kim, *http://orcid.org/0000-0001-5088-2732*
Seong-Doo Hong, *http://orcid.org/0000-0001-8876-7094*

Authors' Contributions

S.G.K. and S.H.S. performed the surgery of this case. S.H.S. wrote the first draft. H.S. revised the first draft and wrote the manuscript. S.G.K. reviewed and revised the manuscript. S.D.H. performed histological analysis and wrote parts of the manuscript. All authors read and approved the final manuscript.

Acknowledgements

This work was performed with the support of "Cooperative Research Program for Agriculture Science and Technology Development (project no. PJ01121404)", Rural Development Administration, Republic of Korea.

Ethics Approval and Consent to Participate

This case report was approved by the Institutional Review Board (IRB) of Gangneung-Wonju National University Dental Hospital, Gangneung, Korea (IRB 2017-002).

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

References

1. Choi YS, Han GS, Choi JS, Jang TY. Two cases of malignant melanoma in turbinate and maxillary sinus. Eur Arch Otorhinolaryngol 2006;263:996-1000.
2. Norhafizah M, Mustafa WM, Sabariah AR, Shiran MS, Pathmanathan R. Mucosal malignant melanoma of the maxillary sinus. Med J Malaysia 2010;65:218-20.
3. Thompson LD, Wieneke JA, Miettinen M. Sinonasal tract and nasopharyngeal melanomas: a clinicopathologic study of 115 cases with a proposed staging system. Am J Surg Pathol 2003;27:594-611.
4. Prasad ML, Busam KJ, Patel SG, Hoshaw-Woodard S, Shah JP, Huvos AG. Clinicopathologic differences in malignant melanoma arising in oral squamous and sinonasal respiratory mucosa of the upper aerodigestive tract. Arch Pathol Lab Med 2003;127:997-1002.
5. Bekci T, Aslan K, Günbey HP, Incesu L. Primary malignant mucosal melanoma of the nasopharynx: an unusual cause of unilateral hearing loss. J Craniofac Surg 2014;25:e567-9.
6. Cheng YF, Lai CC, Ho CY, Shu CH, Lin CZ. Toward a better understanding of sinonasal mucosal melanoma: clinical review of 23 cases. J Chin Med Assoc 2007;70:24-9.
7. Clifton N, Harrison L, Bradley PJ, Jones NS. Malignant melanoma of nasal cavity and paranasal sinuses: report of 24 patients and literature review. J Laryngol Otol 2011;125:479-85.
8. Pathma L, Philip R, Harvinder S, Manjit S. Sinonasal malignant melanocytic melanoma: a rare case report. Med J Malaysia 2008;63:152-3.
9. Rinaldo A, Shaha AR, Patel SG, Ferlito A. Primary mucosal melanoma of the nasal cavity and paranasal sinuses. Acta Otolaryngol 2001;121:979-82.
10. Prasad ML. Update on pigmented lesions of the sinonasal tract. Head Neck Pathol 2007;1:50-4.
11. Roth TN, Gengler C, Huber GF, Holzmann D. Outcome of sinonasal melanoma: clinical experience and review of the literature. Head Neck 2010;32:1385-92.
12. Mochel MC, Duncan LM, Piris A, Kraft S. Primary mucosal melanoma of the sinonasal tract: a clinicopathologic and immunohistochemical study of thirty-two cases. Head Neck Pathol 2015;9:236-43.
13. Som PM, Brandwein-gensler MS, Kassel EE, Genden EM. Tumors and tumor-like conditions of the sinonasal cavities. In: Som PM, Curtin HD, eds. Head and neck imaging. St. Louis: Mosby; 2011:291-2.
14. Kung B, Deschenes GR, Keane W, Cunnane M, Jacob-Ampuero MP, Rosen M. Paranasal sinus melanoma masquerading as chronic

sinusitis and nasal polyposis. Ear Nose Throat J 2007;86:561-4.
15. Giotakis EI, Kotrotsos IN. Inflammatory pseudotumour of the maxillary sinus mimicking malignancy. B-ENT 2015;11:147-50.
16. Snyder ML, Paulino AF. Melan-A as a useful diagnostic immunohistochemical stain for the diagnosis of primary sinonasal melanomas. Head Neck 2002;24:52-5.
17. Narasimhan K, Kucuk O, Lin HS, Heilbrun LK, Carron M, Venkatramanamoorthy R, et al. Sinonasal mucosal melanoma: a 13-year experience at a single institution. Skull Base 2009;19:255-62.
18. Lund VJ, Howard DJ, Wei WI, Cheesman AD. Craniofacial resection for tumors of the nasal cavity and paranasal sinuses: a 17-year experience. Head Neck 1998;20:97-105.
19. Kim CM, Park MH, Yun SW, Kim JW. Treatment of pathologic fracture following postoperative radiation therapy: clinical study. Maxillofac Plast Reconstr Surg 2015;37:31.
20. Kim MK, Han W, Kim SG. The use of the buccal fat pad flap for oral reconstruction. Maxillofac Plast Reconstr Surg 2017;39:5.