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Solitary fibrous tumor in the maxillary sinus treated by endoscopic medial maxillectomy

Satoru Kodama*, Keigo Fujita, Masashi Suzuki

Department of Otolaryngology, Oita University Faculty of Medicine, 1-1 Idaigaoka, Hazama-machi, Yufu, Oita 879-5593, Japan

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Abstract

Solitary fibrous tumor (SFT) is an uncommon neoplasm that usually arises from the pleura. Recently, SFTs have been reported in the head and neck area. SFTs of the nasal cavity and paranasal sinuses are extremely rare, with only 24 cases reported in the English literature. We describe an SFT that arose from the left maxillary sinus and extended to the nasal cavity. The tumor was removed by endoscopic medial maxillectomy, which permitted monobloc excision of the lesion. The patient is without the evidence of the disease 12 months after surgery. © 2008 Elsevier Ireland Ltd. All rights reserved.

Keywords: Solitary fibrous tumor; Maxillary sinus; Endoscopic sinus surgery; Endoscopic medial maxillectomy; Harmonic scalpel

1. Introduction

Solitary fibrous tumor (SFT) is an uncommon neoplasm also known as benign fibrous mesothelioma or submesothelial fibroma; it is one of the different types of mesothelial tumor. SFT was first described in 1931 as a primary spindle cell tumor of the pleura [1] and was later referred to as SFT of the pleura and peritoneum with no actual mesothelial differentiation. The majority of these tumors originate in the pleura, but SFTs can also derive from other serosal membranes. Due to its mesenchymal origin. SFTs have been described in a wide variety of extrapleural locations such as the abdomen, extremities, and vulva [2]. Recently, SFTs have been reported in the head and neck area, parapharyngeal space, tongue, larynx, and parotid gland [3,4]. SFTs of the nasal cavity and paranasal sinuses are extremely rare, with only 24 cases reported in the English literature to date [5,6]. The main treatment for SFT is complete surgical excision. Herein, we describe an SFT that arose from the left maxillary sinus and extended to the nasal cavity that was successfully treated by endoscopic medial maxillectomy (EMM).

2. Case report

The patient was a 74-year-old man with a left nasal tumor that caused progressive left nasal obstruction. Seven years before, a left nasal polyp was found by another otolaryngologist, and the patient underwent endoscopic sinus surgery (ESS) under local anesthesia. The left nasal tumor filled the left middle meatus and originated in the left maxillary sinus. The part of the mass that was in the nasal cavity was removed; however, the base of the mass was left in the maxillary sinus because the surgeon lacked the instruments and experience to perform the more complicated surgery. Postoperative pathological examination revealed SFT. A Caldwell-Luc procedure was offered to the patient to facilitate complete removal of the tumor, but he refused the surgery at that time. The left nasal obstruction gradually worsened, and he was subsequently referred to our hospital for surgery. Endoscopy revealed a pinkish smooth mass that filled the left middle meatus (Fig. 1a). The tumor bled easily when probed with an instrument. Enhanced axial computed tomography (CT) showed a homogeneously enhancing mass that filled the posterior part of the left nasal cavity and the maxillary sinus. No bone damage was observed (Fig. 2a). Magnetic resonance imaging (MRI) showed a left nasal mass with a hypointense

Corresponding author. Tel.: +81 97 586 5913; fax: +81 97 549 0762.
E-mail address: satoruk@med.oita-u.ac.jp (S. Kodama).

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Fig. 1. Endoscopic views of the nose: (a) preoperative view; (b) intraoperative view; (c) postoperative view. (*) The tumor, (**) nasal septum, (***) inferior turbinate, and (****) posterior wall of the maxillary sinus. The base of the tumor is seen in on the medial wall of the left maxillary sinus (b). The maxillary sinus mucosa recovered, and no recurrence was observed (c).

signal on T1-weighted images and heterogeneous hypo and hyperintense signals on T2-weighted images. The mass showed prominent and inhomogeneous enhancement after Gadolinium (Gd) injection. MRI revealed that the tumor originated in the medial wall of the left maxillary sinus (Fig. 2b and c). EMM, which was selected on the basis of the preoperative radiologic findings, was performed under general anesthesia. A sphenopalatine block was performed, and the lateral nasal wall was infiltrated with 1% lidocaine with 1:200,000 epinephrine. Middle meatal antrostomy was performed after complete removal of the uncinate process. An incision was made through the anterior third of the inferior turbinate with the turbinate scissors and biting forceps. The lacrimal bone was cut, and the lacrimal sac and nasolacrimal duct were identified by pressing the left eye. The tumor invasion to the nasolacrimal duct was not shown, and the wall of the nasolacrimal duct was preserved with a safe margin. The anterior portion of the medial maxillary wall was incised with a chisel to make the anterior cut, and an inferior cut was made along the junction between the nasal floor and the floor of the maxillary sinus. The lateral wall was mobilized medially by progressive dissection, allowing entry into the maxillary sinus. The base of the tumor was clearly identified on the medial maxillary wall (Fig. 1b). The harmonic scalpel (HS), an ultrasonic dissector coagulator, was used to excise the mucosa with tumor with the involvement, along with a margin of macroscopically normal mucosa. A posterior cut was made with the HS through the maxillary sinus. The posterior attachment of the inferior turbinate was also cut with the HS, and the medial maxillary wall along with the tumor was successfully removed in one piece. Intraoperative blood loss was less than 5 ml. The tumor size was $40 \text{ mm} \times 32 \text{ mm} \times 18 \text{ mm}$, and the tumor weight was about 22 g (Fig. 3a). Histopathologically, the lesion consisted of spindle-shaped cells with varying amounts of collagen between there. The spindle cells showed a patternless arrangement within the collagenous matrix. Numerous thick-walled vessels and dilated vascular spaces were present (Fig. 3b and c). Immunohistochemically, the tumor cells stained positively for CD34, with no staining for S-100 protein, Bcl-2, or c-kit, thus helping us establish a diagnosis of SFT. The postoperative course was uneventful, and no recurrence was observed at the 1-year follow-up examination (Fig. 1c).

3. Discussion

SFT is a well-recognized entity occurring in the serosal surfaces and most commonly at the level of the pleura [1,2]. SFT occurs more frequently in adults of all ages than in children. Histologically, SFTs are formed by plump spindle



Fig. 2. Diagnostic images of the tumor. (a) Enhanced axial CT shows a homogeneously enhancing mass that fills the posterior part of the left nasal cavity and the maxillary sinus. (b and c) Gd-enhanced axial and horizontal MRI show a prominent and inhomogeneously enhancing mass that originates in the medial wall of the left maxillary sinus.



Fig. 3. Macroscopic (a) and microscopic (b and c) features of the excised tumor. (b) The spindle cells are arranged in a patternless fashion in the collagenous matrix $(100\times)$. (c) Numerous thick-walled vessels and dilated vascular spaces are present $(100\times)$.

cells arranged in a patternless fashion in a collagenous background. Typically, there are hyper and hypocellular areas and prominent vascularity within the lesion that result in a hemangiopericytoma-like pattern. Immunohistochemical assessment permits differentiation of SFT from other fibrous or spindle-cell neoplasms of the upper respiratory tract such as hemangiopericytoma, angiofibroma, fibrous histiocytoma, schwannoma, leiomyoma, fibromatosis, and fibrosarcoma [7]. Recently, CD34, a transmembrane glycoprotein found on the surface of hematopoietic progenitor cells, has been considered a positive marker for SFT. Although CD34 is not a specific marker for SFT, its expression, in conjunction with lack of expression of other immunohistochemical markers, can be helpful in excluding a variety of other soft tissue tumors [7].

Surgery is the treatment of choice for SFT, although radiotherapy and chemotherapy have also been used [8]. SFTs of the head and neck are mostly benign and can be successfully treated with surgical excision. Various surgical procedures, such as lateral rhinotomy, medial maxillectomy, ethmoidectomy, and surgery with a transfacial approach, have been performed [6]. ESS is now widely accepted and commonly performed in cases requiring nose or paranasal sinus surgery. ESS provides superior magnification, illumination, and angled visualization, thereby allowing the surgeon to isolate the base of the tumor and accurately define the extent of disease. ESS for SFT was first described in 2003 [6], and five cases of SFT of the nasal cavity and paranasal sinuses treated successfully by ESS have been reported in the literature (Table 1). To our knowledge, the case reported here is the sixth and the first to include resection by EMM that was completed without complication. EMM was recently validated as an effective treatment for benign sinonasal neoplasm such as inverted papilloma [13,14]. Removing the medial wall of the maxillary sinus via endoscopic approach, with additional removal of the medial buttress and/or lacrimal bone as needed, results in a recurrence rate comparable to that of traditional open medial maxillectomy. The advantages of EMM are lack of facial or gingival incision, lack of external scar, no loss of bony nasal or anterior maxillary support, low risk of infraorbital nerve paresthesia, short hospital stay, less postoperative pain and facial swelling, less blood loss and crusting, and possibly less epiphora [14]. Morbidity is also low. In addition to patient's comfort, a systematic review of the literature supports EMM as a favorable treatment option for most cases of benign sinonasal tumor, revealing lower recurrence rate compared to external approaches [15,16]. In the case described herein. EMM was shown to be an effective treatment for maxillary SFT.

Maxillary sinus involvement constitutes a dilemma in the management of the tumor. The assessment of endoscopic resectability depends on preoperative endoscopic findings and imaging. The skill of the surgeon is also an important factor when selecting a surgical technique [13]. Surgeons with extensive training and experience in ESS would likely be more comfortable in utilizing endoscopic techniques as extent of the disease increases. Tumors originating from the

Table 1							
Reported	solitary	fibrous	tumor	cases	treated	by	ESS

reported somming notous tamor eases abated of 200								
Author (year)	Age and sex	sex Site ^a Size (mm) ESS ^b		Follow-up (months)				
Alobid I (2003) [6]	43 M	Rt, E, NC	65 imes 38 imes 30	Ethmoidectomy, sphenoidectomy	12			
Pasquini E (2003) [9]	54 F	Rt, E, NC	?	Ethmoidectomy, sphenoidectomy	18			
Eloy PH (2006) [10]	26 F	Rt, E, NC	50 imes 35 imes 28	Ethmoidectomy, middle turbinectomy	6			
Corina (2006) [11]	63 F	Rt, E	?	Ethmoidectomy, sphenoidectomy	6			
Sciarretta V (2006) [12]	73 F	?, E, NC	?	Ethmoidectomy	12			
Kodama S (2008) ^c	74 M	Lt, M, NC	$40\times32\times18$	Medial maxillectomy, ethmoidectomy	12			

^a Rt, right side; Lt, left side; E, ethmoid; M, maxillary; NC, nasal cavity.

^b Endoscopic sinus surgery.

^c The present case.

maxillary sinus eroding the medial wall of the sinus and extending into the nose may be managed through an endoscopic approach if the surgeon thinks that adequate visualization is possible endoscopically and that the tumor can be totally resected in this manner. The limits of EMM are the posterior wall of maxillary sinus posteriorly, the orbital floor superiorly, the floor of the nose inferiorly, and the anterior wall of the maxillary sinus anteriorly. If any question exists regarding the ability to dissect to the periphery of the maxillary sinus in a manner sufficient to remove the entire tumor with clear mucosal margins, then the procedure can be converted into an open, extranasal procedure with a medial maxillectomy or Caldwell-Luc approach [16].

Control of intraoperative bleeding is very important in ESS. SFTs are often hypervascular and can bleed easily [7]. In fact, failure of ESS to successfully remove SFT due to massive intraoperative bleeding has been reported [17,18]. A new surgical tool, the HS, cuts and coagulates simultaneously by means of ultrasound technology, and there have been many reports of the efficacy of the HS in surgery [19,20]. Recently, we reported the effectiveness of HS for excision of bleeding polyps in the nasal cavity. Blood loss was minimal, and operation time was short [21]. In the case described herein, the HS was useful for the excision of the SFT; it eliminated bleeding and did not delay wound healing.

In conclusion, ESS can be successfully used to achieve the complete removal of benign tumors such as SFTs that arise at the level of the nasal cavity and paranasal sinus. In comparison to traditional approaches, ESS has several advantages, including no need for external incision, decreased blood loss, low morbidity, decreased hospital stay, and possibility of repetition in cases of recurrence. EMM may become the treatment of choice for maxillary SFT.

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References

[1] Klemperer P, Rabin CB. Primary neoplasms of the pleura. A report of five cases. Arch Pathol 1931;11:385–412.

- [2] Nielsen GP, O'Connell JX, Dickersin GR, Rosenberg AE. Solitary fibrous tumor of soft tissue: a report of 15 cases, including 5 malignant examples with light microscopic, immunohistochemical, and ultrastructural data. Mod Pathol 1997;10:1028–37.
- [3] Witkin GB, Rosai J. Solitary fibrous tumor of the upper respiratory tract. A report of six cases. Am J Surg Pathol 1991;15:842–8.
- [4] Safneck JR, Alguacil-Garcia A, Dort JC, Phillips SM. Solitary fibrous tumor of two new locations in the upper respiratory tract. J Laryngol Otol 1993;107:252–6.
- [5] Zukerberg LR, Rosenberg AE, Randolph G, Pilch BZ, Goodman ML. Solitary fibrous tumor of the nasal cavity and paranasal sinuses. Am J Surg Pathol 1991;15:126–30.
- [6] Alobid I, Alos L, Blanch JL, Benitez P, Bernal-Sprekelsen M, Mullol J. Solitary fibrous tumour of the nasal cavity and paranasal sinuses. Acta Otolaryngol 2003;123:71–4.
- [7] Mentzel T, Bainbridge TC, Katenkamp D. Solitary fibrous tumour: clinicopathological, immunohistochemical, and ultrastructual analysis of 12 cases arising in soft tissues, nasal cavity and nasopharynx, urinary bladder and prostate. Virchows Arch 1997;430:445–53.
- [8] Goodlad JR, Fletcher CD. Solitary fibrous tumour arising at unusual sites: analysis of a series. Histopathology 1991;19:515–22.
- [9] Pasquini E, Cantaroni C, Salfi N, Tamburini G, Marchi C, Sciarretta V. Endoscopic treatment of an ethmoidal solitary fibrous tumour. J Laryngol Otol 2003;117:889–91.
- [10] Eloy PH, Nollevaux MC, Watelet JB, Van Damme JP, Collet ST, Bertrand B. Endonasal endoscopic resection of an ethmoidal solitary fibrous tumor. Eur Arch Otorhinolaryngol 2006;263:833–7.
- [11] Corina L, Volante M, Carconi M, Contucci AM. An unusual solitary fibrous tumor after sphenoethmoidectomy. Otolaryngol Head Neck Surg 2006;134:1063–5.
- [12] Sciarretta V, Pasquini E, Frank G, Modugno GC, Cantaroni C, Mazzatenta D, et al. Endoscopic treatment of benign tumors of the nose and paranasal sinuses: a report of 33 cases. Am J Rhinol 2006;20:64–71.
- [13] Wormald PJ, Ooi E, van Hasselt CA, Nair S. Endoscopic removal of sinonasal inverted papilloma including endoscopic medial maxillectomy. Laryngoscope 2003;113:867–73.
- [14] Sadeghi N, Al-Dhahri S, Manoukian JJ. Transnasal endoscopic medial maxillaectomy for inverting papilloma. Laryngoscope 2003;113:749– 53.
- [15] Busquets JM, Hwang PH. Endoscopic resenction of sinonasal inverted papilloma: a meta-analysis. Otolaryngol Head Neck Surg 2006;134:476–82.
- [16] Krouse JH. Endoscopic treatment of inverted papilloma: safety and efficacy. Am J Otolaryngol 2001;22:87–99.
- [17] Kessler A, Lapinsky J, Berenholz L, Sarfaty S, Segal S. Solitary fibrous tumor of the nasal cavity. Otolaryngol Head Neck Surg 1999;121:826–8.
- [18] Morales-Cadena M, Zubiaur FM, Alvarez R, Madrigal J, Zarate-Osorno A. Solitary fibrous tumor of the nasal cavity and paranasal sinuses. Otolaryngol Head Neck Surg 2006;135:980–2.
- [19] Sugo H, Mikami Y, Matsumoto F, Tsumura H, Watanabe Y, Kojima K, et al. Hepatic resection using the harmonic scalpel. Surg Today 2000;30:959–62.
- [20] Kunde D, Welch C. Ultracision in gynaecological laparoscopic surgery. J Obstet Gynaecol 2003;23:347–52.
- [21] Kodama S, Yoshida K, Nomi N, Fujita K, Suzuki M. Successful treatment of nasal septum hemangioma with the harmonic scalpel. A case report. Auris Nasus Larynx 2006;33:475–8.