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Case Report

### A rare pediatric variant of organized hematoma in the maxillary sinus



Young Ah Cho<sup>a,1</sup>, Ik Jae Kwon<sup>b,1</sup>, Soung Min Kim<sup>b,\*</sup>, Hoon Myoung<sup>b</sup>, Jong Ho Lee<sup>b</sup>, Suk Keun Lee<sup>c</sup>

- <sup>a</sup> Department of Oral and Maxillofacial Pathology, School of Dentistry, Kyung Hee University, Seoul, Republic of Korea
- b Department of Oral and Maxillofacial Surgery, Dental Research Institute, School of Dentistry, Seoul National University, Seoul, Republic of Korea
- <sup>c</sup> Department of Oral Pathology, College of Dentistry, Gangneung-Wonju National University, Gangneung, Republic of Korea

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#### ABSTRACT

Organized hematoma of the maxillary sinus is an uncommon, benign, heterogeneous, non-neoplastic, and hemorrhagic lesion characterized by mucosal swelling and bone destruction that can be locally aggressive in the unilateral maxillary sinus. The clinical, radiographical, and pathological characteristics of a 2-year-old patient with an organized hematoma in the maxillary sinus were studied with preoperative computed tomography (CT) and magnetic resonance imaging (MRI) scans and histopathological examination with hematoxylin and eosin (H&E), masson trichrome (MT), and immunohistochemical stainings of vascular markers, such as CD31, factor VIII, and smooth muscle actin (SMA); and lymphatic factors, such as D2-40; and proliferation factors, such as Ki-67 and bcl-2.

A total of 17 articles of similar pathology have been reported in the English-language literature under the name of an organized hematoma in the maxillary sinus. We reviewed these retrospective studies of 60 patients who were treated surgically within the past 17 years and found no consensus on the diagnostic criteria of this disease or surgical approaches. Patient age ranged from 11 to 78 years, and the average age was 42.8 years. The ratio of male to female was approximately 2:1, no cases in pediatric patients were found. We report the first case of organized hematoma in the pediatric maxillary sinus with its clinico-radio-pathologic characteristics. Preoperative imaging was essential for the accurate diagnosis of this disease, and sublabial approach combined with endonasal endoscopic confirmation can be also recommended in the pediatric patient.

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#### 1. Introduction

Organized hematoma of the maxillary sinus is an uncommon, benign, heterogeneous, non-neoplastic, and hemorrhagic lesion characterized by mucosal swelling and bone destruction that can be locally aggressive in the unilateral maxillary sinus. This lesion was first reported as a tumor in the Japanese literature in 1917 by Tadokoro as a "blood boil of the maxillary sinus." It is now well known in Japan and Korea as part of the differential diagnosis

for maxillary sinus tumors. Hemangioma in the maxillary sinus was also reported before the 1990s, but terms such as 'organized hematoma,' 'hematoma in the maxillary sinus,' or 'organized hematoma mimicking tumor in the maxillary sinus' have been reported more recently [1-3].

Hematoma, the accumulation of hemorrhage, often results in the impairment of body function, depending on the involved organ and the amount of bleeding. Some critical conditions, such as hemoperitoneum, hemothorax, and hemopericardium, are well-known sequelae of hemorrhage into a body cavity [4,5]. Such accumulation of hemorrhage also occurs in the sinonasal cavity, though it is not life threatening. Unlike hemorrhage in other body cavities, this stagnant blood in the sinus cannot be drained quickly and is usually found to be coagulated and organized [4,6]. In addition, because it can mimic other sinus pathology by expanding into adjacent tissues and displacing or destroying them, the antral organized hematoma must be differentiated from sinusitis, mucocele, antral polyp, cholesterol granuloma, inverted papilloma, vascular tumors, and malignant neoplasms [6,7].

<sup>☆</sup> Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

<sup>\*</sup> Corresponding author at: Department of Oral and Maxillofacial Surgery, School of Dentistry, Seoul National University, 101 Daehak-ro, Jongno-gu, 110-768 Seoul, Republic of Korea. Tel.: +82 2 2072 0213; fax: +82 2 766 4948.

E-mail addresses: smin5@snu.ac.kr, smin\_kim@msn.com (S.M. Kim).

<sup>&</sup>lt;sup>1</sup> These authors contributed their own works as the first author.

**Fig. 1.** (A) Panoramic radiograph shows slight posterior displacement of a tooth germ of the right maxillary first molar (*white arrows*). (B) An enhanced CT image reveals a non-enhancing mass occupying the right maxillary sinus. The mass protrudes into the retroantral space through the posterolateral wall of the right maxillary sinus (*black arrowheads*). (C) An axial, fat-suppressed, T2-weighted MR image demonstrates a heterogeneous signal intensity mass. (D) Markedly high signal intensities (*black arrows*) indicating a hematoma are observed within the mass on an axial T1-weighted MR image.

Over the past 17 years, a total of 17 articles describing similar pathology have been reported in the English-language literature under the name of organized hematoma in the maxillary sinus, with no consensus on diagnostic criteria or surgical approaches for treatment. After review of 60 patients in 17 articles, no cases in pediatric patients were found. The present study aims to report the first case of organized hematoma arising in the maxillary sinus of a 2-year-old infant and to review the literature of organized hematoma cases from 1996 to 2013 with analysis of the clinical, radiologic, and pathologic features.

#### 2. Case report

#### 2.1. Pediatric patient evaluation and surgery

A 2-year-old boy was referred to the Department of Oral and Maxillofacial Surgery at Seoul National University Hospital, Seoul, Korea. This patient suffered from nasal congestion, coryza, cough, and sore throat for two weeks. Because a blood clot was found in his throat during physical exam in a private clinic, a panoramic radiograph was performed and revealed a right maxillary lesion with displaced tooth germs (Fig. 1A). He was referred to our center for further evaluation and treatment.

Clinically, a firm swelling in the right upper buccal vestibule was observed with slight facial swelling. The first upper premolar was immobile and visualized with good occlusion state. The second premolar had not yet erupted. Endoscopic nasal examination showed bloody rhinorrhea without purulence, and the middle meatus with the uncinate process could be seen with slightly edematous nasal mucosa. The boy had no medical history, no allergies, and no previous hospitalizations. His young parents are not smokers.

Enhanced computed tomography (CT) revealed a  $2.9\,\mathrm{cm} \times 2.0\,\mathrm{cm} \times 2.6\,\mathrm{cm}$ , multilocular mass occupying the sinus cavity (Fig. 1B). The lateroposterior wall of the maxillary sinus was deformed, and the temporal muscle was compressed by the mass.

The tooth germ of the right maxillary first molar was displaced with partial loss of its bony crypt. While T2-weighted magnetic resonance images (MRI) showed heterogeneous signals that indicated a mixture of fluid and soft tissue (Fig. 1C), T1-weighted MR images demonstrated high signal intensities in the mass (Fig. 1D). Therefore, the radiological findings were more consistent with hematoma/hemorrhage, vascular anomaly, or aneurysmal bone cyst than a malignant tumor. Based on the nature and duration of the disease with clinical and radiographic findings, the tentative preoperative diagnosis was hematoma in the maxillary sinus, mesenchymal sarcoma, ameloblastoma, or dentigerous cyst.

Whole-mass excision was performed under general anesthesia via sublabial approach combined with endonasal endoscopic surgery. A careful subperiosteal dissection was performed for full exposure of the involved maxillary wall (Fig. 2A). Arterio-venous bleeding from the inner-medial wall of the maxillary sinus was coagulated with vessel clamp and electrocautery (Fig. 2B) after removal of the mass. The gross morphology of the excised mass and teeth looked like a benign mass with a thick fibrous cystic wall.

During 20 months of follow-up, the patient has not had any sinus problems or recurrence, as demonstrated by radiographic and endonasal sinuscopic examinations.

## 2.2. Immunohistochemical study to detect CD31, CD34, factor VIII, SMA, and D2-40

The biopsy specimens were fixed in 10% buffered formalin solution, embedded in paraffin, and sectioned to 4 µm thickness. Microsections were stained with hematoxylin and eosin, and immunohistochemical analysis was performed using the streptavidin–biotin–peroxidase complex method as previously described [8]. Primary antibodies for analysis included vascular markers, such as CD31, CD34, factor VIII, and smooth muscle actin (SMA); lymphatic factors, such as D2-40; and proliferation factors, such as Ki-67 and bcl-2. Antibodies were diluted to 1:100

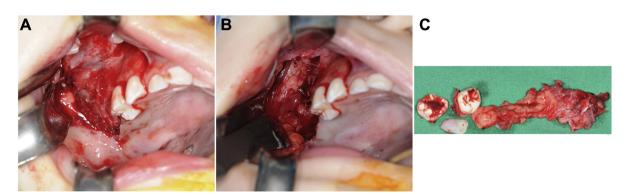


Fig. 2. Intraoperative clinical views: (A) intraoral mass before excision, (B) removal state, and (C) mass with teeth after removal.

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