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Case report

En bloc resection of intraosseous arteriovenous malformation (AVM) in the maxilla and deformed bone following embolization and sclerotherapy

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ABSTRACT

Introduction: Arteriovenous malformations (AVMs) of facial bones are rare conditions. These lesions can cause aesthetic challenges, pain, impact on gums and teeth, facial asymmetry, and life-threatening bleeding.
Presentation of the case: This report described a case involving the resection of AVMs within the maxilla following preoperative embolization and sclerotherapy by using an ultrasonic bone aspirator (Sonopet®, Stryker, Kalamazoo, MI, USA) and a radiofrequency bipolar sealer (Aquamantys™, Medtronic, Minneapolis, MN, USA). The AVMs and deformed bone were resected en bloc through repeated cutting and hemostasis, preserving the maxillary crown and maintaining the maxillary structure by treating only the affected bone.
Discussion: Reducing inflow during surgery and controlling intraoperative bleeding requires thorough planning. Preoperative embolization and sclerotherapy can reduce inflow to the tumor, and appropriate surgical devices enable the resection of arteriovenous malformations within the maxilla.
Conclusion: The method presented in this report is a viable treatment option because it allows for selective bone removal, thus leading to the preservation of crowns and maxillary contour.

1. Introduction

The occurrence of arteriovenous malformations (AVMs) in facial bones is rare, with the mandible and maxilla being the most frequently affected bones [1,2]. The characteristic features of bony AVMs of the maxilla or mandible include spontaneous bleeding (often minor and recurrent), tooth mobility, and gingival discoloration. Another typical symptom of intraosseous AVMs is progressive bone deformity, which can lead to asymmetry and aesthetic compromise [2]. Although endovascular treatments and surgical resection have been reported, treating these lesions is often challenging. The treatment of intraosseous vascular malformations carries a high bleeding risk and can be difficult. Even with preoperative embolization and sclerotherapy, controlling bleeding during surgical resection can be challenging [2]. To control intraoperative bleeding and maintain a clear surgical field, we performed resection using an ultrasonic bone aspirator (Sonopet®, Stryker, Kalamazoo, MI, USA) and a radiofrequency bipolar sealer (Aquamantys™, Medtronic, Minneapolis, MN, USA). This report aimed to describe a case in which a maxillary AVM, along with the deformed bone, was resected following preoperative embolization and sclerotherapy. This method allowed for the resection of only the affected area and the preservation of the maxillary tooth roots. Although a defect was observed in the anterior wall of the maxillary sinus, it was still possible to preserve the buttress and contour-influencing parts. This work has been reported in line with the SCARE criteria [3].

2. Presentation of the case

The patient was a 44-year-old male with a history of AVM in the left preauricular region, which was previously treated with sclerotherapy. Approximately one year ago, he noticed a mass in the left cheek in addition to the known preauricular AVM. The mass gradually enlarged and became painful, prompting him to present at our department. A hard, immobile mass measuring approximately 9 cm × 7 cm was noted in the left cheek, along with a soft, mobile mass measuring approximately 5 cm × 4 cm in the preauricular region. Ultrasonography revealed a fast-flowing mass. Computed tomography (CT) imaging showed that the left maxilla was thinned and protruded, with a contrast-enhanced tumorous lesion inside (Fig. 1). The preauricular AVM was confined to the soft tissue without bone involvement. A partial biopsy

Abbreviations: AVM, arteriovenous malformation; CT, computed tomography; 3D, three-dimensional.

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revealed a benign hemangioma. The patient initially requested the resection of only the mass in the left cheek. Considering the risk of increased bleeding, we also decided to proceed with the resection of only the lesion. Suspecting an AVM within the maxilla, we attempted resection following preoperative coil embolization by the radiology department. However, significant bleeding occurred when the intra-osseous portion of the lesion was reached. Clamping the external carotid artery did not control the bleeding, with an estimated blood loss of 500 mL within a short period. Anticipating further hemostatic difficulties, we decided to terminate the surgery without resection. The surgical plan was subsequently revised. Preoperative embolization and sclerotherapy were performed, and appropriate surgical devices were selected.

Two days before the surgery, embolization and sclerotherapy were performed on the inflow vessels (Fig. 2). Following coil embolization,

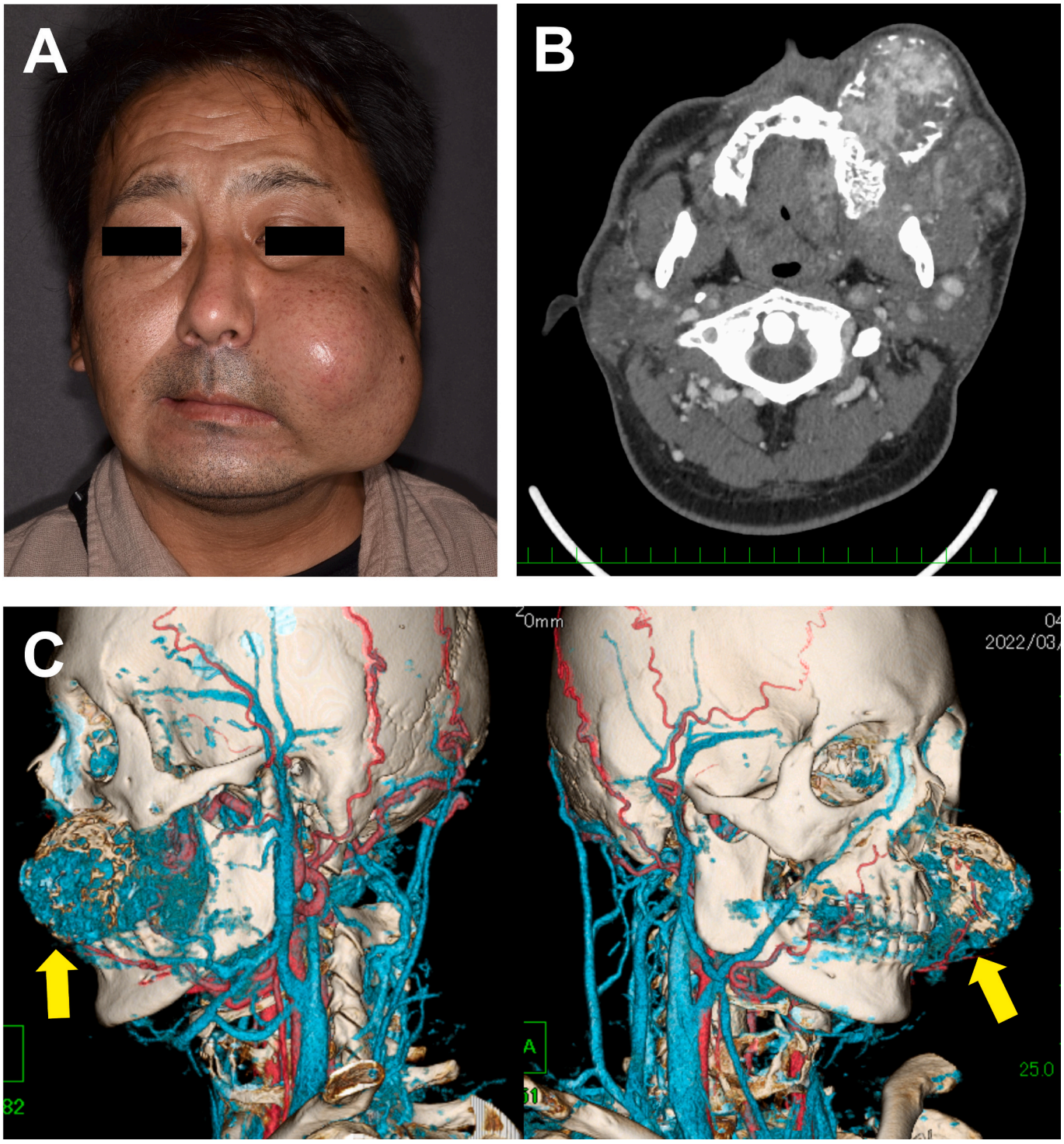

Fig. 1. (A) Clinical findings: Two prominent masses were observed in the left cheek and preauricular area. (B) Enhanced CT imaging: Contrast-enhanced CT images showed a thinned and protruding left maxilla with a contrast-enhanced mass lesion inside. (C) 3D CT imaging: The anterior wall of the left maxilla was deformed and significantly protruding because of the tumor (yellow arrow). Abnormal venous vessels were also observed on the bone surface.
CT, computed tomography; 3D, three-dimensional.

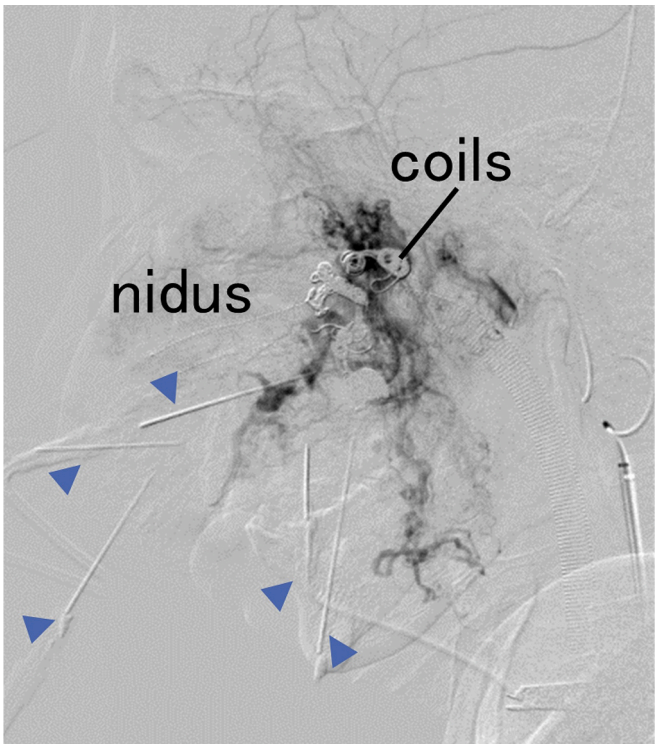

Fig. 2. Two days before the surgery, coil embolization and sclerotherapy with a 1:3 polidocanol–air solution (indicated by blue arrows) were performed.

polidocanol foam sclerotherapy was administered. The surgery was performed using a Weber–Ferguson incision. The entire lesion was exposed by subperiosteal dissection. Additionally, the infraorbital nerve was encased in the vascular malformation, ligated, and divided. By using the Sonopet, we excised the margins of the thinned bone lesion. Thereafter, by using the Aquamantys for coagulation and hemostasis, we repeatedly performed cutting and coagulation to resect the AVM en bloc (Fig. 3). This method allowed for the preservation of the maxillary tooth roots. Although a defect was observed in the anterior wall of the maxillary sinus, it was still possible to preserve the buttress and contour-influencing parts. Intraoperative blood loss was 2730 mL, which required a transfusion of 4 units (560 mL) of packed red blood cells. Postoperatively, no additional bleeding or worsening of anemia was observed, and no further transfusions were required. Hence, the histopathological diagnosis of the specimen was AVM. Sensory numbness in the infraorbital nerve region and mild incomplete paralysis in the zygomatic branch of the facial nerve (with the ability to forcibly close the caudal orbicularis oculi) were observed; however, these symptoms gradually improved. Eight months postoperatively, the appearance of the left cheek had improved. The surgical scar has healed without being noticeable. To further improve the aesthetic outcome, we are considering the removal of the preauricular AVM in the future.

3. Discussion

Most extracranial AVMs occur in the head and neck regions, with the maxilla and mandible being the most commonly affected bones [1]. Intraosseous AVMs of the facial bones present with symptoms, such as pain, facial asymmetry, swelling, bleeding, tooth mobility, gingival discoloration, and bone deformity [2]. Maxillary AVM treatment must consider bleeding control, bone management, function, occlusion, and contours. The reported treatment options for intraosseous AVMs include selective embolization, sclerotherapy, and surgical resection. Endovascular procedures offer the advantage of being minimally invasive. Conversely, surgical removal can result in functional or aesthetic impairments. From a radical treatment perspective, some researchers argue that surgical treatment following selective embolization is the best option [2]. Surgical resection of intraosseous AVMs of the facial bones can be complicated by significant bleeding.

Azzolini et al. [4] described a method involving tooth extraction, removal of vascular malformations through the interalveolar bony septa, cavity scraping, and packing with absorbable hemostatic material after endovascular treatment. However, this method can result in tooth loss. Brusati et al. [5] reported a technique involving selective arterial embolization, hole creation in the cortical bone with a burr, curettage of the lesion with a small spoon, and immediate packing of the holes with oxidized cellulose. Behnia et al. [6] described two methods: (1) segmental mandibulectomy with intraosseous lesion curettage, extraction of the involved teeth, and immediate reconstruction of the cortical bone as well as (2) intraosseous lesion curettage after proximal mandibular osteotomy. Additionally, Colletti et al. [2] reported a method involving aggressive curettage and packing with regenerated cellulose 24 h following preoperative embolization with n-butyl cyanoacrylate. However, they noted a major drawback of curettage: severe bleeding that was difficult to control. The same report stated that maxillary AVMs are more likely to occur because the palatal cortex is thinner than the mandibular lingual cortex. Similarly, they mentioned cases in which massive intraoperative bleeding prevented the radical resection of large maxillary AVMs, even with preoperative embolization and partial external carotid artery clamping during surgery. Treatment methods should be considered on the basis of the anatomical aspects and patient preferences for each case. In this case, we performed a combination of selective coil embolization and sclerotherapy with polidocanol to reduce the AVM blood flow as much as possible, followed by resection surgery. Selective arterial coil embolization was performed to reduce the inflow, and sclerotherapy was conducted to prepare for venous bleeding. Despite preoperative embolization and sclerotherapy, the AVM remained as a high-flow lesion, thus necessitating the use of surgical devices, such as Aquamantys and Sonopet, to control intraoperative bleeding. The effectiveness of Aquamantys for hemostasis in bone and soft tissue has been well documented. Aquamantys is a radiofrequency bipolar hemostatic sealer that combines radiofrequency energy with saline to achieve hemostasis in soft tissues and bones during surgery, thereby reducing blood loss. Operating at approximately 100 °C, it provides desirable effects on surrounding tissues owing to its low temperature and allows the occlusion of vessels <1 mm in diameter [7]. Moreover, it is used in joint replacements, spinal surgeries, and hepatic and pancreatic resections, with reported reductions in blood loss and transfusion requirements [8–10].

Sonopet is an ultrasonic surgical device that simultaneously performs fragmentation, aspiration, and irrigation. Its vibrating tip at 25 KHz directly contacts and fragments the tissue, which is then aspirated. Non-rotational reciprocating motion allows for bone cutting without entanglement or kicking, and irrigation ensures thermal safety. It is widely used in the craniofacial, neurosurgical, and spinal fields, thus demonstrating its effectiveness and minimal invasiveness into the surrounding tissues [11].

In this case, we used Aquamantys for hemostasis to control bleeding and maintain the surgical field. Similarly, Sonopet was used to manage the enlarged and thinned bone, minimizing damage to the AVM and surrounding soft tissues. The AVM and deformed bone were resected en bloc by alternating between cutting and hemostasis. This approach allowed for selective bone removal, preserving the crowns and maintaining the maxillary contour.

4. Conclusion

We used Aquamantys for hemostasis to control bleeding and maintain the surgical field. Similarly, Sonopet was used to manage the enlarged and thinned bone, minimizing damage to the AVM and surrounding soft tissues. The AVM and deformed bone were resected en

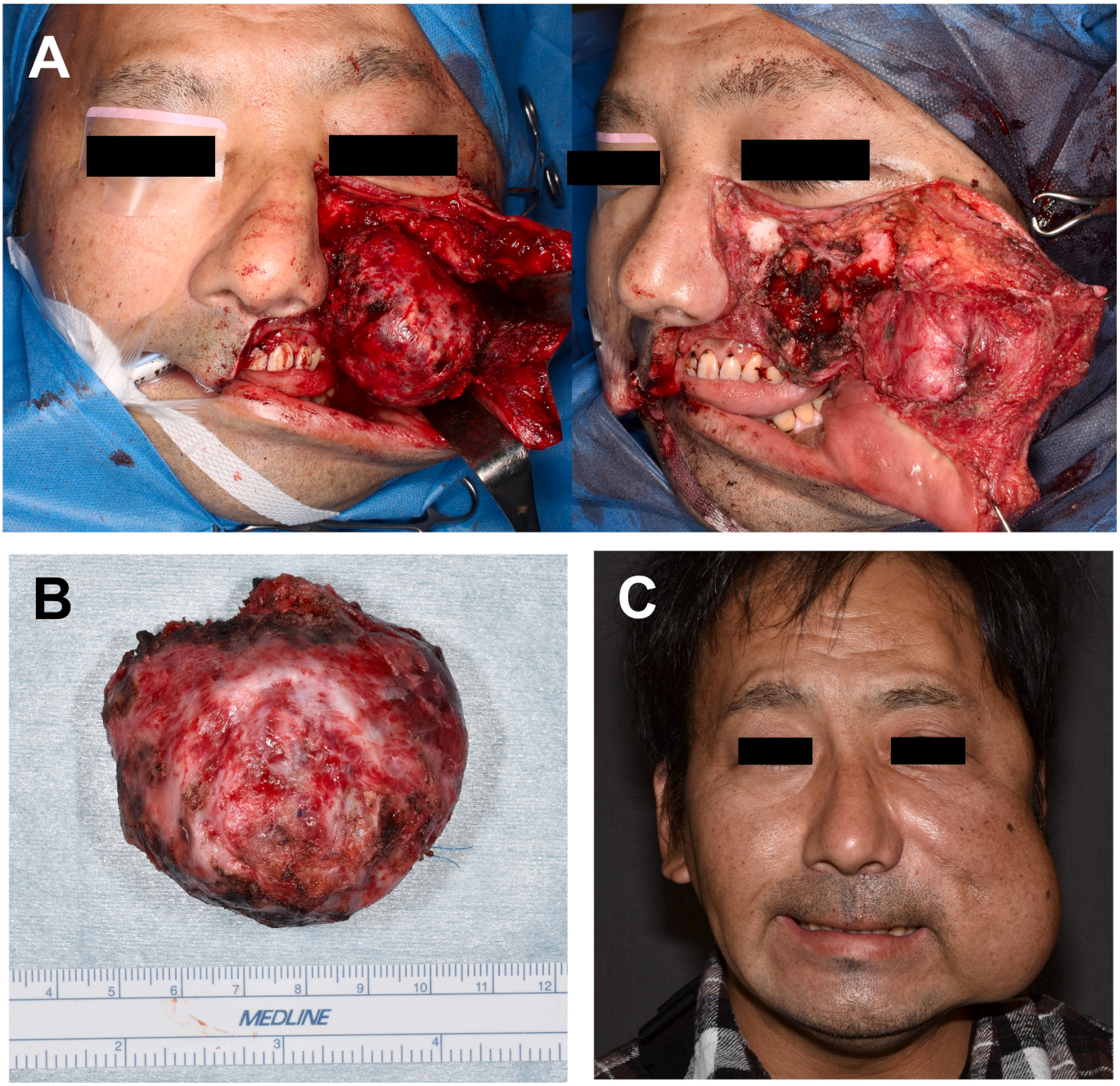

Fig. 3. (A) The approach was made via a Weber–Fergusson incision, fully exposing the tumor. The AVM was excised in one piece along with the deformed bone while alternating between cutting and hemostasis. (B) Resected specimen. (C) Clinical photograph eight months after surgery. The protrusion of the left cheek has improved. The AVM in front of the ear is scheduled for excision in the future.
AVM, arteriovenous malformation.

bloc by alternating between cutting and hemostasis. This approach allowed for selective bone removal, thus preserving the crowns and maintaining the maxillary contour.

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Author contribution

Misato Ueda; writing—original draft preparation, review and editing.
Tadashi Nomura; writing – review & editing.
Shunsuke Sakakibara, Hiroto Terashi; supervised the work.
All authors have agreed to the published version of the manuscript.

Registration of research studies

Not applicable.

Consent

Written informed consent was obtained from the patient or their parents for publication of this case report and accompanying images. A

copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This is a case report. Thus, this study was exempt from institutional review board approval.

Guarantor

Misato Ueda.

Declaration of competing interest

None.

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