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Case Report

Arteriovenous malformation on the sole of the foot treated successfully by embolization ☆,☆☆

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ABSTRACT

Arteriovenous malformations of the sole of the foot are rare and can cause disturbances in normal living activities. We report a case of a plantar arteriovenous malformation in a 24-year-old male with pain and difficulty in walking. The arteriovenous malformation was complex, with a large and poorly marginated nidus, so we considered that with surgical resection, walking disabilities would be inevitable. When surgical removal of vascular mass is difficult, embolization alone can be effective. Therefore, he was treated with 4 therapeutic embolization procedures. Transvenous approaches to the venous sac and direct punctures of the nidus was performed. The nidus was successfully eradicated by embolization using alcohol, resulting in the disappearance of associated symptoms. Appropriate imaging is essential for diagnosis and evaluation of treatment. We were successful in achieving improved quality of life and satisfaction for a rare and difficult case by percutaneous embolization and sclerotherapy.

Abbreviations: AA, Arterial Aneurysm; AVF, Arteriovenous Fistula; AVM, Arteriovenous Malformation; CT, Computed Tomography; CTA, Computed Tomography Angiography; DSA, Digital Subtraction Angiography; ECT, Enhanced Computed Tomography; EO, Ethanolamine Oleate; HHT, Hereditary hemorrhagic telangiectasia; MRA, Magnetic Resonance Angiography; MRI, Magnetic Resonance Imaging.

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Introduction

Arteriovenous malformations (AVMs) are fast-flow abnormal connections between arteries and veins, bypassing the capillary system. The treatment goal for AVMs is eradication of the nidus to improve symptoms, but surgical excision is difficult because most AVMs infiltrate into normal tissue. Therefore, embolization based on arteriographic classifications is useful to eradicate the nidus. AVMs of the sole of the foot are rare and can cause disturbances in normal living activities. Here, we report a case of a patient with an AVM on the sole of the foot, treated successfully by embolization.

Case report

A 24-year old male with an enlarged throbbing painful mass on the left sole of the foot, noticed from the age of 8, was admitted to our hospital. On physical exam, a 5-cm diameter lesion was pulsating on the left plantar in the first metatarsal phalangeal joint (Fig. 1). The epidermal color and texture were within normal range. Enhanced computed tomography (ECT) showed abnormal tortuous vessels and early visualization of the venous sac and draining veins in the first and second proximal phalangeal area (Fig. 2). Digital subtraction angiography (DSA) displayed the medial plantar and dorsal pedis arteries as thickened, tortuous, and connected to the nidus (Fig. 3). The nidus had numerous small collaterals, where some shunted into a dilated venous sac (type II shunts based on Cho et al.'s classification [1]). Multiple shunts were also found between arterioles and venules (type IIIb shunts).

Four sessions of treatment were performed in total. In the first session, transarterial sclerotherapy was performed (Fig. 4). The popliteal artery was temporarily occluded using a 5.2 Fr balloon catheter (Serecon MP catheter; Terumo Clinical Supply Co., Ltd., Gifu, Japan). Then, a microballoon catheter (Logos; Piolax, Yokohama, Japan) was advanced coaxially to the distal portion of medial plantar and dorsal pedis artery. Using a tourniquet on the thigh for additional blood flow control, a 5% foam sclerosant (a mixture of 10% ethanolamine oleate [Takeda, Osaka, Japan] and the same amount of nonionic contrast material and CO_2) was injected into the nidus. DSA showed a remarkably decreased blood flow in the nidus. However, 2 days after the procedure, blisters and purpura emerged on the skin surrounding the AVM, leading to an ulcer (Fig. 4). Prostandin (Alprostadil Alfadex) ointment was applied. After 5 months of outpatient follow-up, the wound had healed. On DSA, the proximal feeding arteries and a part of the nidus had occluded. However, proliferation of innumerable new feeding vessels had also occurred.

On the second and third procedure, the draining vein was punctured and a 1.9 Fr microcatheter alone was inserted into the venous sac of the nidus (type II component) and ethanol was injected for eradication.

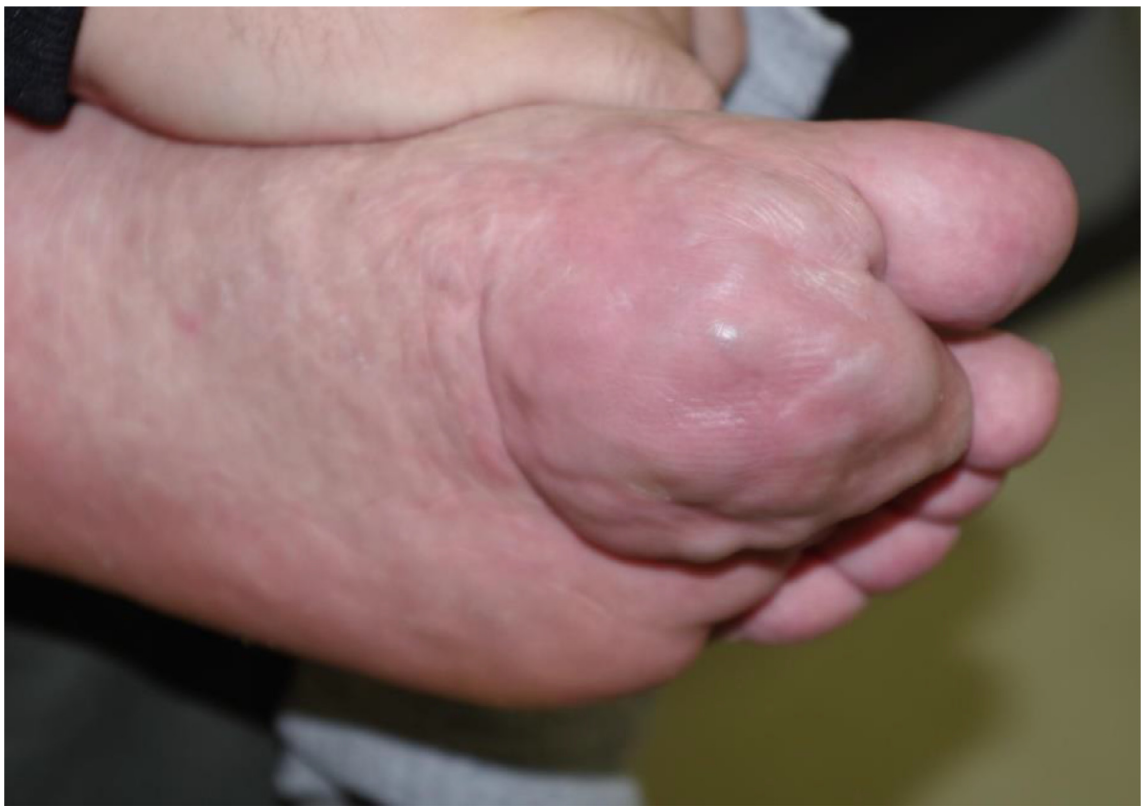

Fig. 1 – A throbbing painful mass on the left sole of the foot at first visit. A pulsating 5-cm diameter mass was observed in the first metatarsal phalangeal joint. The epidermal color and texture were within normal range.

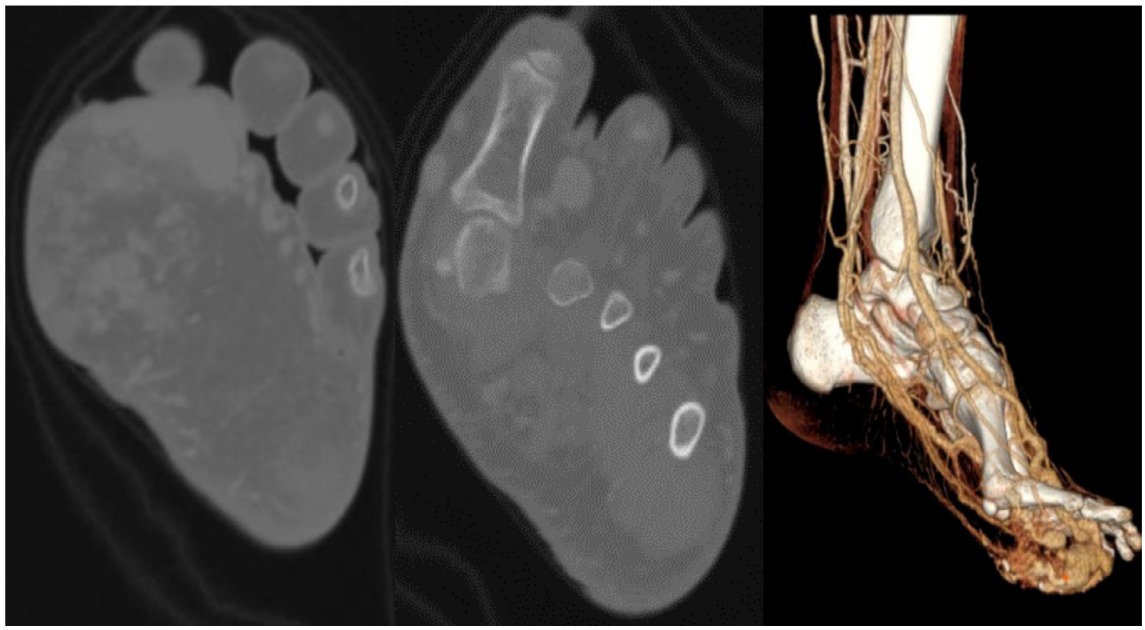

Fig. 2 – Enhanced computed tomography scan with volume rendering. The medial planter and dorsal pedis arteries, thickened and tortuous, shunting randomly with surrounding veins. The draining veins were visualized earlier through the nidus, as abnormal wiry vessels, and as venous sacs around the first and second proximal phalangeal area.

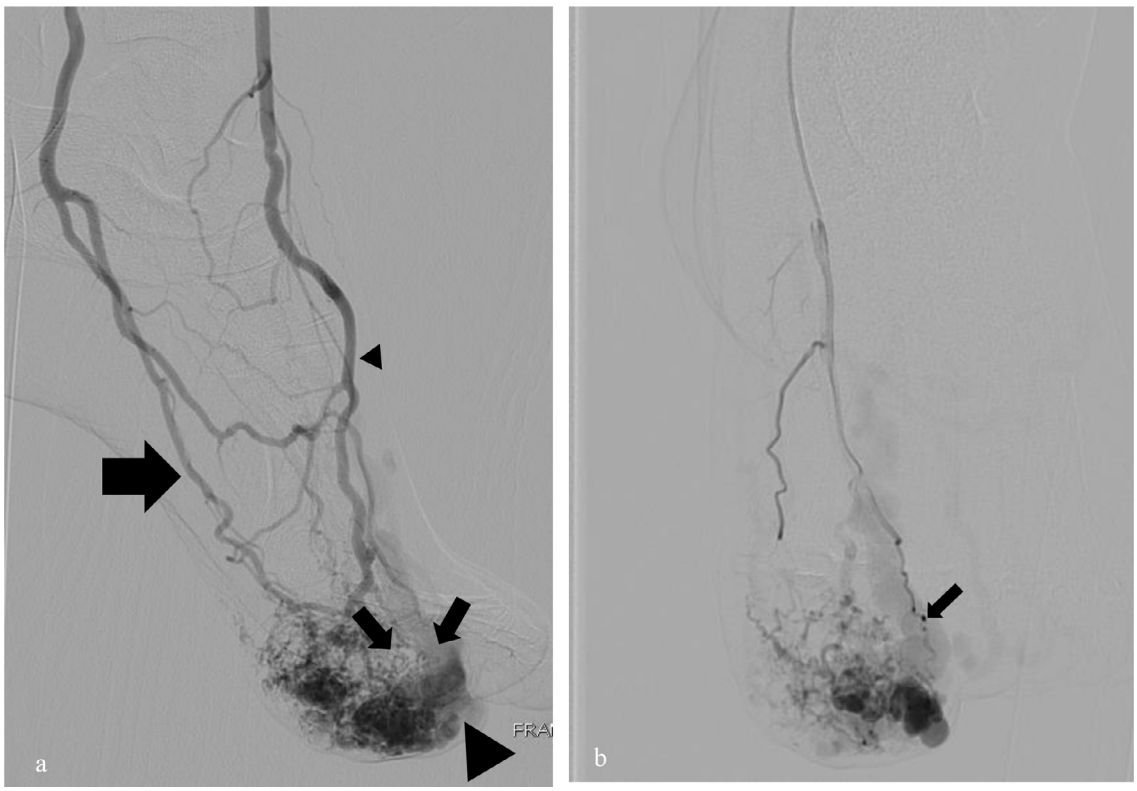

Fig. 3 – Digital substraction angiography before treatment, (a) showing multiple fine afferent arteries (small arrow) originating from the medial plantar (large arrow) and dorsal pedis artery (small arrowhead), shunting to the venous sac (large arrowhead) (type II). (b) Multiple fine arteries connected directly to fine veins (arrow) (type III).

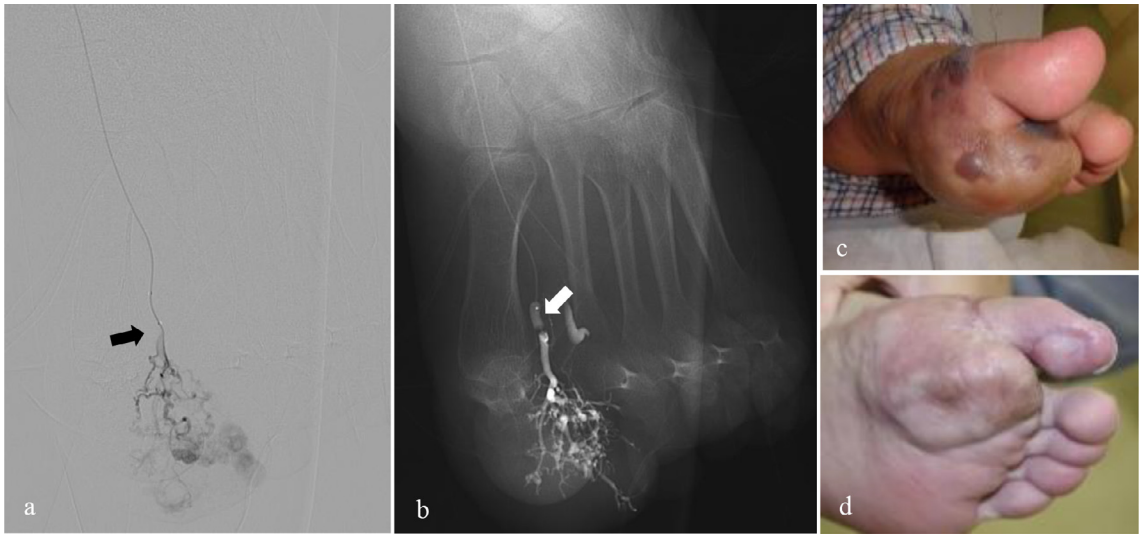

Fig. 4 – The first procedure and its complications. (a) Digital subtraction angiography in arterial phase. Contrast agent diluted into the distal part of medial plantar artery (arrow). (b) Sclerosant injected into the nidus under balloon inflation (arrow). (c) Blisters and purpura emerged at 2 postoperative days. (d) The ulcer showed complete epithelialization after 5 postoperative months.

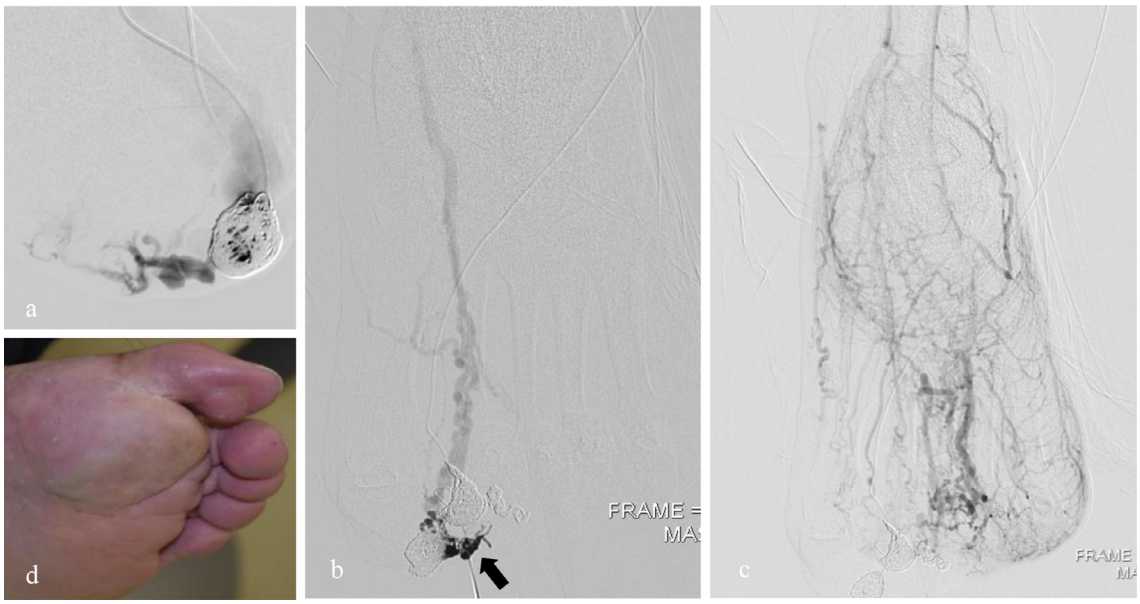

Fig. 5 – DSA and photograph after all 4 procedures. (a) Venous sac embolized with coils, followed by alcohol and/or NBCA injection. (b) Direct puncture of the type III component (arrow), followed by alcohol injection. (c) Final DSA showing complete eradication of the nidus. (d) Three years from the fourth session. The pulsation disappeared, and the mass shrunk and is painless and soft. He was able to wear shoes and walk freely with no symptoms.

On the fourth procedure, the venous sac was embolized using metallic coils to stagnate the nidus flow, followed by injection of ethanol mixed with contrast material (8:2) under blood flow control of the popliteal artery, similar to the first session (Fig. 5). For the type IIIB component, direct puncture of the tortuous vessels connected to the draining veins was performed, followed by repeated injection of 80% alcohol.

After 4 sessions, DSA showed eradication of the nidus, and on physical exam, the pulsation disappeared, and the mass shrunk (Fig. 5). The patient's pain completely disappeared, and he was able to wear shoes and walk freely with no symptoms for his follow-up period of 3 years from the last embolization.

Discussion

AVMs are fast-flow abnormal connections between arteries and veins, bypassing the capillary system. They may form due to the failure of regression of arteriovenous channels in the

primitive retiform plexus [2]. The population prevalence is approximately 10 per 100,000 in the United States [3], and they most commonly affect the head and neck (47.4%) but are also found in the extremities (28.5%) [4]. AVMs occur with equal frequency in males and females, and 40%-60% of lesions are visible at birth, and 30% become clinically apparent during childhood [2]. Having a family history of AVMs may rarely increase the risk of developing AVMs, but most types are not inherited. Certain hereditary conditions such as hereditary hemorrhagic telangiectasia may also increase the risk of AVMs [5]. For prognosis, between 40% and 80% of patients report improvement in symptoms after successful embolization and cure is achieved in 10% or fewer of cases [6]. Recurrence is common (up to 50%), particularly in diffuse AVMs affecting the extremities [6].

On physical exam, peripheral AVMs present in a wide variety of location and size. They usually protrude as a pulsating mass and can be redder in comparison to the surrounding skin. Local symptoms vary and include tissue overgrowth, local hypervascularity, steal phenomenon, hyperemia, pain, bleeding, ischemia, and ulcers. General symptoms include venous hypertension and even high-output cardiac failure [7].

For diagnosis, ultrasound (US), computed tomography (CT), and magnetic resonance imaging (MRI) are commonly used noninvasive studies for peripheral lesions. US examination reveals multiple vascular channels with feeding arteries, nidus, and draining veins seen as multiple anechoic spaces [8]. On CT, AVM is delineated as a nonspecific soft tissue attenuation of mass. CT angiography displays feeding arteries as hypertrophied and serpentine, the nidus as tangled vessels, and the draining veins as enlarged and early filling. The venous phase displays equilibration of enhancement in arteries and veins [9]. MRI and MR angiography are used to define the morphology of these lesions, which typically demonstrate hypointense tubular or nodular flow voids on both T1 and T2 sequences with lack of mass effect or mass enhancement. Fatty hypertrophy and muscular atrophy may also be seen [8]. Bone involvement in AVMs is best demonstrated on postcontrast T1-weighted images with intensive contrast uptake of the intraosseous vessels [10].

On angiography, AVM shows tortuous and dilated arteries with arteriovenous shunting, nidus, and enlarged draining veins [11]. Angiography is used to define the main arterial supply to the vascular malformation, the presence of a nidus, the size of arteriovenous shunting, and the venous drainage [12], for both diagnosis and consideration of treatment.

AVMs of the sole of the foot are rare and can cause disturbances in normal living activities [13,14]. Surgical excision of the AVM was the gold-standard treatment but is difficult because AVMs usually exist inside normal tissue, where the loss of their normal function is inevitable; in this case, it would mean a partial foot amputation. However, surgical ligation or coil embolization of the feeding artery is also difficult and possibly harmful due to consequent development of numerous collateral feeders shunting to the nidus [14,15]. This nidus is referred to as the first dilated segment of vein after the abnormal connections between arteries and veins [16]. The treatment goal for AVMs is eradication of the nidus to improve symptoms and prevent recurrence [16–18].

To plan the optimal approach, predict therapeutic outcomes, and minimize non-target embolization of peripheral AVMs, classification by angiography is used. Various angiographic classifications of the nidus have been proposed [1,13]. The present case used Cho et al's classification and included type II and IIIb lesions. Cho et al classified peripheral AVMs into 4 types; type I: arteriovenous fistulae, type II: arteriolovenous fistulae, type III: arteriolovenulous fistulae with nondilated (IIIa) or dilated (IIIb) fistulae or combinations thereof [1]. They reported best therapy outcomes in patients with Type II AVM after accessing this type typically through direct puncture or transvenous approaches with various agents [1]. In our case, for the type II lesions, the venous sac was cannulated and embolized using coils to achieve stagnation of flow, followed by alcohol injection. For the type IIIb lesions, alcohol was injected by direct puncture of the nidus. The results were satisfactory, with symptoms alleviating and no gait impairment.

The rate of reported complications, such as tissue necrosis and neuropathy ranges from 10% to 30% [19]. Injection of ethanol into a normal artery can cause severe tissue necrosis by thrombosing and destroying nutritive capillary beds. Cho et al reported that skin necrosis is the most frequent complication in sclerotherapy using alcohol. All their cases of skin necrosis or bullae healed by using only topical wound dressings [1]. In our first session, ethanolamine oleate was transarterially injected. Although normal arteries were not visualized on DSA, extensive skin necrosis occurred. Stagnation of the sclerosant outside the nidus and in the draining vein may result in extensive skin necrosis.

Yakes et al reported 3 cases of plantar AVMs treated by sclerotherapy [13]. They used alcohol as the main sclerosant to obliterate the nidus. Alcohol (50%) mixed with contrast was transarterially injected into the nidus and pure alcohol was used for direct puncture. All their cases were curative with minor skin necrosis in only two. The treatment of type IIIa AVMs seems difficult because only transarterial access is available [1].

Conclusion

Percutaneous embolization and sclerotherapy based on the vascular architecture of AVMs, appears to be a safe and effective method in eradicating the nidus and curing AVMs.

Ethics in publishing

Approved by the Institutional Review Board (IRB) – Institution: Keio University, National committee: Independent Ethics Committee, IRB #20160049.

Human and animal rights

No experiments on human or animal subjects were performed.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Submission declaration and verification

This article has not been previously published in any form.

Use of inclusive language

All writing in this article is free from bias, stereotypes, slang, reference to dominant culture and/or cultural assumptions.

Authors contribution

Ikki Yuzaki: Drafting of manuscript; Noriko Aramaki-Hattori: Study conception and design; Masashi Tamura: Assistant in interventional procedure; Hideyuki Torikai: Assistant in interventional procedure; Keisuke Okabe: Clinical patient work; Shigeki Sakai: Analysis and interpretation of data; Seishi Nakatsuka: Analysis and interpretation of data; Kazuo Kishi: Critical revision; Masanori Inoue: Interventional procedure; All authors have read and approved the final version of the manuscript.

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Informed consent and patient details

Informed consent and consent for publication was obtained for every individual person's data included in the study.

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