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Spitzoid melanoma of the finger: a case report

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Abstract

Background Melanoma is the most malignant skin tumor, with a high metastatic potential.

Spitzoid melanoma is a subtype of melanoma requiring rapid management and extensive tumor resection.

We have set the goal to recognize anatomical peculiarities and difficulties diagnoses posed by this type of tumor, as well as to recognize the management modalities, especially the surgical one, of malignant spitzoid melanoma.

Case presentation A 25-year-old Tunisian male patient had consulted for nodular lesion of the right index, evolving for 4 years. A malignant tumor was strongly suspected, then confirmed as a melanoma by a biopsy excision.

Initially, the excision was incomplete in depth, suggesting a complementary surgery, but the patient refused it; 3 years later, the patient again consulted after the appearance of an axillary lymphadenopathy and worsening of the skin lesion. A supplement of tumor removal with lymph node biopsy were performed. It was decided to perform an amputation of the second ray and the first commissure with cheiroplasty, reconstructing a four-finger hand. An homolateral axillary cleaning was performed at the same time.

The postoperative result is considered esthetically and functionally satisfying.

The evolution was marked by the appearance of pulmonary metastases, requiring adjuvant chemotherapy.

A regression of the nodule under the mammary skin and total disappearance of axillary nodes have been marked; but the patient's condition rapidly deteriorated, and he died after a 2-month decline.

Conclusion Spitzoid melanoma is exceptional, posing difficulties in diagnostics, and it should not be underestimated, considering that it may involve the vital prognosis.

Knowledge of this rare form of melanoma is important to avoid misdiagnosis, which delays diagnosis and subsequent therapy.

Keywords Skin, Tumor, Melanoma, Surgery, Reconstruction, Case report

Introduction

Malignant skin tumors are an entity requiring surgical treatment, which can be mutilating because it sometimes demands a wide-ranging excision. This treatment can even lead to an amputation of a limb or a limb segment. Melanoma is the most malignant skin tumor, which has a high metastatic potential. It originates from melanocytes, which are cells that produce melanin, the pigment responsible of the color of the skin, the eyes, and the hair.

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Malignant melanoma accounts for 5% of all skin cancers, and is the leading cause of death from skin cancer, with a 75% incidence rate [1]. Its incidence is constantly increasing in all populations. The high mortality associated with this condition makes early diagnosis very important. The pathological type of tumor and the depth of the invasion are determinants of overall prognosis. Digital melanoma is rare, occurring most often in the elderly, representing less than 2% of all skin melanomas [2]. Spitzoid melanoma is a subtype of melanoma, which makes its diagnosis is difficult. It looks clinically and histologically like a Spitz nevus, with high metastatic potential and a bleak prognosis, requiring fast management and wide tumor resection [3].

Through an observation of a patient operated on at the orthopedic department of Habib Bourguiba Hospital Sfax – Tunisia and a literature review, we have set the goal of:

- Recognizing anatomical peculiarities and difficulties diagnoses posed by this type of tumor.
- Recognizing the management modalities, especially the surgical one, of malignant spitzoid melanoma.

Case presentation

A 25-year-old Tunisian male patient with no specific medical history had consulted for nodular lesion of the right index, evolving for 4 years. This lesion was papular and 5 mm in diameter, increasing gradually in size and becoming painful. Clinical examination at the admission showed a fairly well-limited nodular lesion infiltrated, firm of 2 cm long axis, with ulcerated center and hyperkeratotic area, sitting in look at the radial edge of the metacarpophalangeal joint of the right index finger (Fig. 1). A malignant tumor was strongly suspected.

A node biopsy excision showed a cellular proliferation made of large or fusiform epithelioid cells with little abundant cytoplasm weakly eosinophilic without melanic pigments and with atypical nucleus well nucleated and hyperchromatic; these cells were arranged in small clusters or in high-level theses at contact of the dermoepidermal junction, giving a characteristic spitzoid appearance; mitoses were quite a lot especially in depth and stroma was profuse desmoplastic (Fig. 2).

Immunohistochemical examination showed tumor cells positive for vimentine, protein S100, Melan Aet HMB4, thus confirming the diagnosis of melanoma.

Initially, the excision was incomplete in depth, suggesting a complementary surgery, but the patient refused it and was lost to follow-up, but 3 years later, the patient again consulted after the appearance of an axillary lymphadenopathy and worsening of the skin lesion (Fig. 3). A supplement of tumor removal with lymph node biopsy was performed (Fig. 4). The histological study confirmed

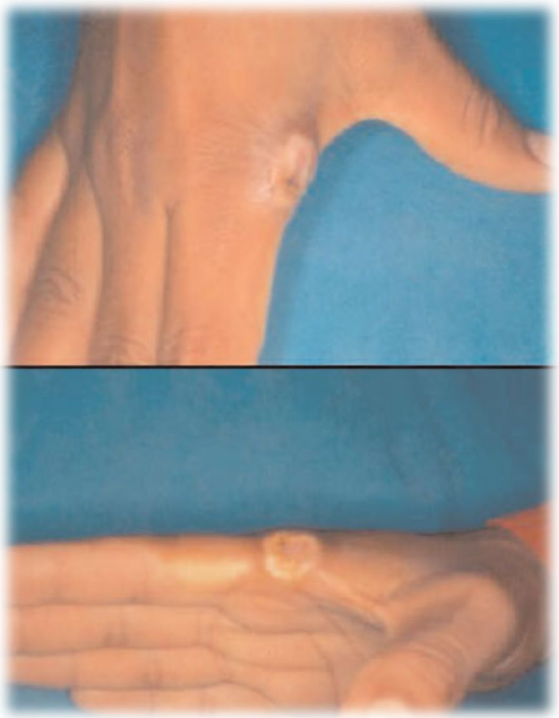

Fig. 1 Ulceroinvasive lesion, 1.5 cm in diameter, next to the radial edge of right index metacarpal-phalangial joint

the presence of a tumor residue whose resection was incomplete (type R1) with axillary ganglionic metastases. It was decided to peform an amputation of the second ray and the first commissure with cheiroplasty, reconstructing a four-finger hand (Figs. 5 and 6). The reconstruction of the first commissure was done by a simple graft of total skin and not by a flap (Fig. 7).

A homolateral axillary cleaning was performed at the same time (Fig. 8).

The pathological examination of the surgical part showed tumor residue in hypodermis, striated muscle, and metastases in lymph nodes (6N+/25N). The Breslow index was estimated at 11 mm, and the tumor was classified pt4 N3 Mx.

Result

The postoperative result was considered esthetically and functionally satisfying.

Evolution was marked by the appearance of pulmonary metastases, a nodule at the junction of the upper quadrant of the right breast, in view of the left upper iliac spine, and a supraclavicular node. Detogenic adjuvant chemotherapy was introduced with a dose of 250 mg/m^2. The patient received three chemotherapy treatments in full. A regression of the nodule under the mammary skin and total disappearance axillary nodes were seen; but the

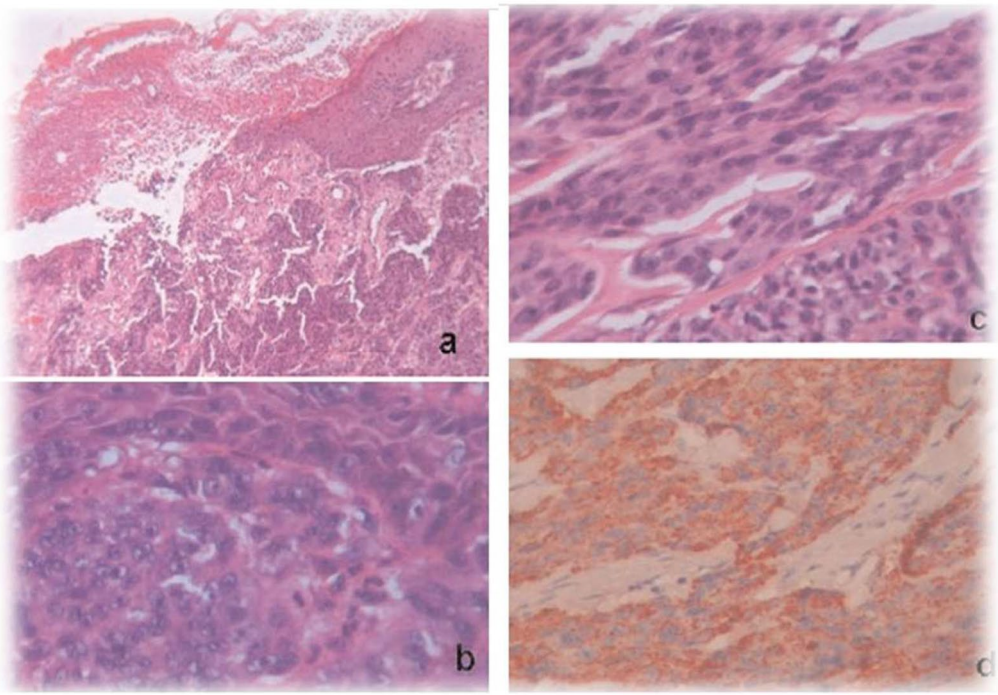

Fig. 2 **a** Tumor proliferation widely ulcerated on the surface (hematoxylin and eosin × 100). **b** Tumor proliferation of oval globular cells in the dermoepidermal junction. Clear artifact spaces separating cells tumors of squamous cells (hematoxylin and eosin × 400). **c** Proliferation of tumor cells infiltrating the deep dermis. Note the presence of prominent nucleoli (hematoxylin and eosin × 400). **d** HMB45 immunopositive (× 400)

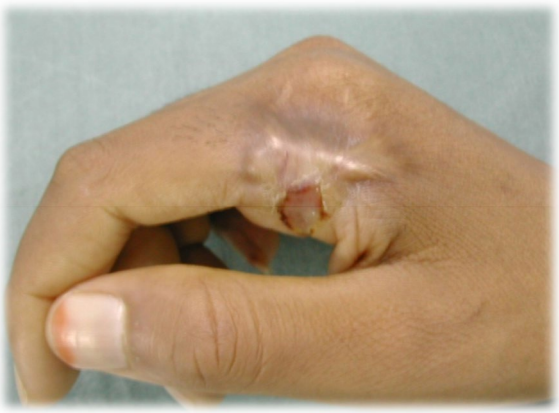

Fig. 3 Clinical aspect of the lesion after 3 years of loss to follow-up

patient's condition rapidly deteriorated, requiring the cessation of chemotherapy and treatment. The patient died after a 2-month decline.

Discussion

Spitz tumors and nevi were first described in 1948 by Spitz as "juvenile melanomas" [4, 5].

Barnhill *et al.* [6] classified Spitz tumors into three categories: tumor of Spitz without atypia (classical Spitz nevus), atypical Spitz tumor, and spitzoid malignant melanoma (SMM). However, in a panel study of 10 pathologists who examined 30 Spitz tumors (including Spitz tumors without or with atypia and spitzoid melanomas), no consensus could be reached on the morphological criteria distinguishing between different types of Spitz tumors [7].

Fewer than 30 spitzoid melanoma observations have been reported in the literature. Spitzoid melanoma is therefore rare; its localization in the finger is exceptional and often poses a diagnostic problem. Spitzoid melanoma can occur in children, but is most common in adults [8–10]. Clinically, spitzoid melanomas are nodular lesions that vary over time, often reaching 1 cm or diameter. The nodules are generally amelanotic. They can look like hemangiomas, pyogenic granulomas, xanthogranulomas, or basal cell carcinomas. Less often, lesions are pigmented and have irregular and variable color. Nodular lesions may be crusty and ulcerated.

In our observation, the lesion was unusual on the one hand by its localization at the finger, and on the other hand by its clinical presentation; in effect, the occurrence of the lesion in a young man and its presentation in the

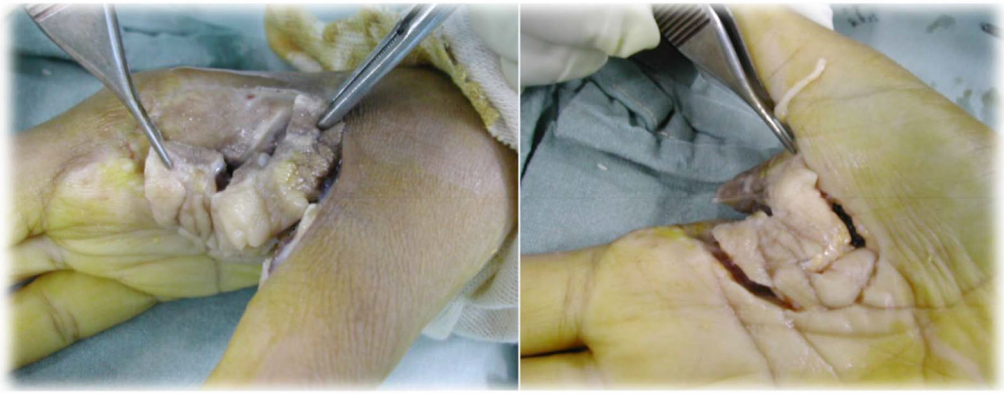

Fig. 4 Tumor removal

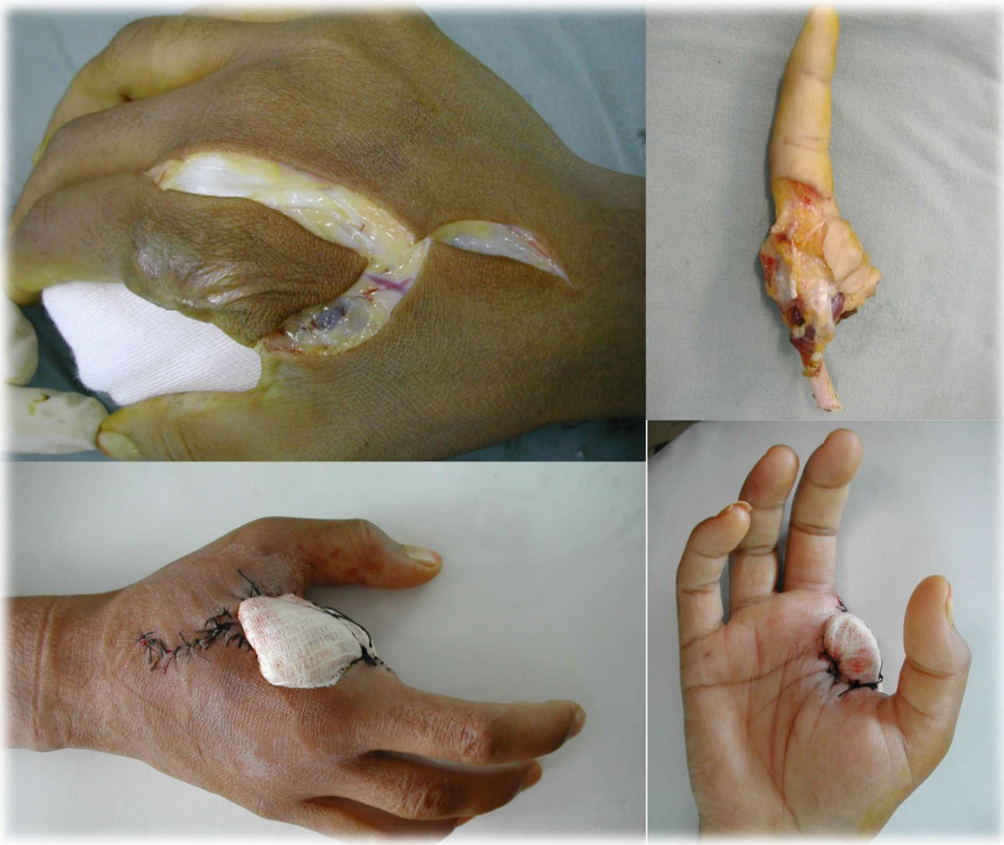

Fig. 5 Amputation of the second radius and first commissure with cheiroplasty

form of an unpigmented infiltrated ulceration did not make suspect a melanoma; the first diagnoses mentioned were sarcoma or lymphoma, but histological examination ruled out these two diagnoses on the morphology of the tumor and confirmed the melanic nature of the lesion due to immunolabeling by PS100, Melan A, and HMB 45.

The main problem of histological differential diagnosis is the challenge in distinguishing between spitzoid melanoma and Spitz nevus [3], especially in young patients. Indeed, spitzoid melanoma is exceptional in young people, therefore, many arguments are required to make this diagnosis.

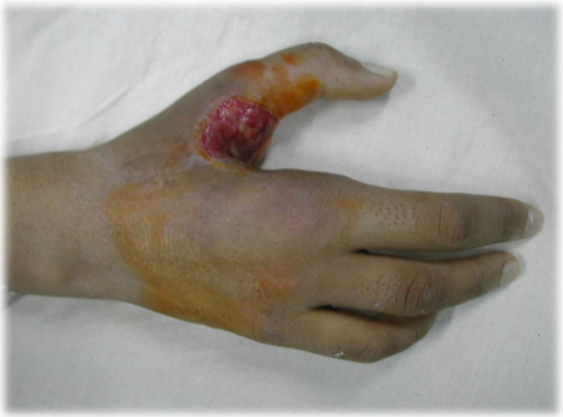

Fig. 6 Reconstruction of a four-finger hand

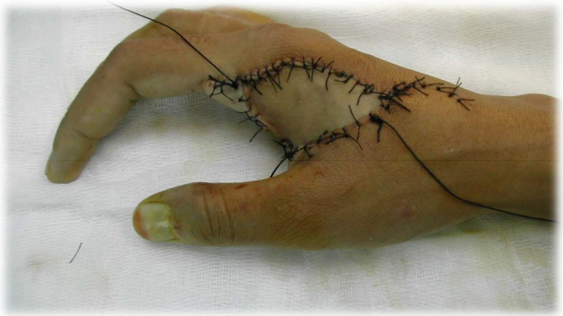

Fig. 7 Reconstruction of the first commissure by total skin graft

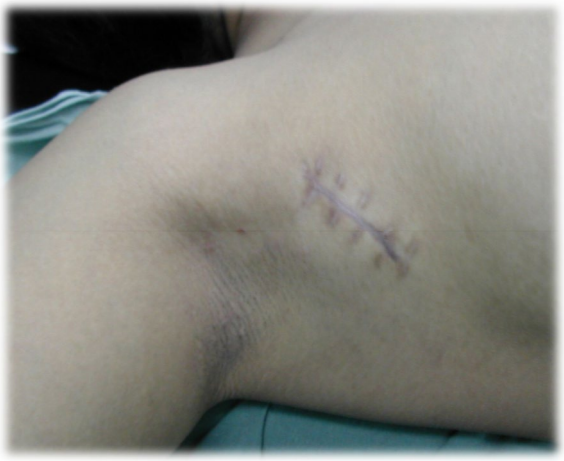

Fig. 8 Axillary removal site

Histopathological features useful for distinguishing the spitzoid melanoma of a Spitz nevus have been reported by several authors, and are represented by the asymmetry of the lesion, ulceration, irregularity of limits, reaching the junction dermoepidermal, gradual decrease in maturation of melanocytes, increased cell density, and a high mitotic index [11–13]; in our case, the ulcerating and especially infiltrating character of the tumor proliferation was evident from the first biopsy, which allowed for retaining the diagnosis of melanoma.

However, in most cases, these features do not allow for distinction between spitzoid melanoma and Spitz nevus, and recently there have been studies that attempt to use molecular cytogenetics for said distinction. Bastian *et al.* [14, 15] demonstrated that 80% of the Spitz with an 11p chromosome have an increased number of copies of an allele HRAS oncogenic; this HRAS allele is rarely mutated into malignant melanoma [3, 14, 15]. Stratification of the risk of the atypical spitzoid melanocytic proliferation was recently addressed with the use of a new test in situ four-probe fluorescence (FISH) hybridization for 6p25 (RREB1), 11q13 (CCND1), 9p21 (CDKN2a), and 8q24 (MYC) [16] and have identified:

- Spitzoid melanoma with homozygous 9p21 deletion (high risk).
- Spitzoid melanoma with gain of 6p25 and/or 11q13 (medium to high risk).
- Atypical Spitz tumor with isolated deletion of 6q23 (risk weak).
- Atypical Spitz tumor without FISH abnormality (low to very high risk weak).

In studies using immunohistochemistry, the proliferation index of Ki-67/MIB-1 was suggested as a useful marker to distinguish nevi from melanoma Spitz [17]. In our case, the tumor cells were positive for vimentine, protein S100, and Mélan Aet HMB45 in the immunohistochemical study.

During histological diagnosis of melanoma, the anatomopathological study of the entire tumor allows for the measurement of the thickness, thus establishing the Breslow index, the main prognostic criterion [18]. However, surgical recovery will often be necessary depending on this histological analysis.

Other major prognostic criteria are provided by the anatomopathological study of the primary tumor with Clark level [19] and the presence or absence of an ulcer, which was the case for our observation.

Finally, this histological study will establish the local stage of melanoma according to the American Joint Committee on Cancer (AJCC) Melanoma Committee classification [20].

Melanoma of the finger has a poor prognosis compared with other localizations of melanoma. Although it is accessible and discovered early, melanoma of the finger is often considered clinically as a benign lesion, delaying

diagnosis and treatment [21]. Average follow-up of melanoma of the finger depends closely on the delay of diagnosis after the appearance of the tumor as well as the anatomical site of the tumor. The subungual melanoma has a poorer prognosis than other melanomas of the finger due to its more aggressive behavior and its fast metastatic dissemination. The 5-year survival is estimated at 37% for ungual localization, and 75% in outside the subungual location [21]. Local recurrence of melanoma is a strong marker of aggressiveness. Survival in case of metastasis distance is 6–8 months [22].

In our observation, the tumor was located at the index distance from the subungual region, however, evolution was rapidly metastatic due to delayed diagnosis.

Management of patients with histopathological diagnosis of a spitzoid melanoma should be decided with a multidisciplinary and informed-consent approach. It follows the same treatment as other types of melanoma, and depends on tumor thickness, Breslow classification, and Clark level.

The opportunity for sentinel node biopsy for melanoma has to be assessed on a case-by-case basis. The decision must be made, whereas in the melanomas of Spitz, the presence of tumor cells isolated in the sentinel node is not a sign of equivocal malignancy [23, 24] and is not an indication of complementary lymphadenectomy, due to ambiguity of the primary syndrome. In our observation, biopsy of an axillary node was done and histological study confirmed lymph node metastasis, leading to a subsequent removal during amputation of the second ray.

For melanoma in situ, excision must have a margin of 0.5 cm of healthy skin. For invasive melanomas less than 1 mm thick, the sound margin must be 1 cm; for 1–2 mm the thickness of the margin is 1–2 cm, and for melanoma 2–4 mm thick, the recommended margin is 2 cm [25–27]. Amputation is still possible and must be done at distance of the lesion when that is circumferential. Although radical amputations are to be avoided even in cases of multiple lesions, they can prove to be the most carcinological surgery, with reassuring results. They are a necessity when the margins of an excision always keep tumor residue, which is the case with delayed diagnosis and therapy.

In our case, an amputation of the second ray was carried out due to the very invasive character of the tumor. We were led to make a cheiroplasty [28, 29] of the hand since the index finger is the most used finger after the thumb. This is an essential element of the pollicidigitale socket and a stabilization in the global digitopalmar grip [30].

Cheiroplasty is called recovery plastie by Tubiana. It may be considered to cover a loss of substance from the hand or a finger. In our clinical case it covered more than half of the loss of dorsal substance. The remaining part was covered by a total skin instead of an associated posterior interbone flap generally done with cheiroplasty [31], given the difficulty of dissection, risk of necrosis, and invasive technique in a field with a metastatic tumor, with a precarious general condition.

Conclusion

Spitzoid melanoma is exceptional, posing difficulties in diagnostics, mainly with the Spitz nevus.

Knowledge of this rare form of melanoma, with its unusual morphology, is important to avoid misdiagnosis as well as delays in diagnosis and subsequent therapy.

Histological and immunohistochemical studies contribute to the diagnostic confirmation of spitzoid melanoma. Suspect criteria in this entity are large asymmetric lesion, high cell density clone, epithelioid or atypical cytology, multiple and deep mitosis, and focal rate of proliferation. In front of a spitzoid histological aspect of a lesion in a young subject, it is important not to underestimate a spitzoid melanoma, which may involve the vital prognosis in case of delayed diagnosis or therapy.

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Author contributions
All authors were involved in writing, reading, and editing the manuscript. All authors read and approved the final manuscript.

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Data availability
Data supporting the findings of this study are available from the corresponding author upon reasonable request.

Declarations

Ethics approval and consent to participate
Ethical approval is not required at our institution to publish an anonymous case report.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

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