Dermatology Reports
https://www.pagepress.org/journals/index.php/dr/index

eISSN 2036-7406

Please cite this article as:

Ciccarese G, Bortone R, Meduri AR, et al. Morbihan disease in an Asian patient: diagnostic and therapeutic challenges. *Dermatol Rep 2026 [Epub Ahead of Print] doi: 10.4081/dr.2026.10487*

Received: 24 June 2025; Accepted: 23 April 2026.

Morbihan disease in an Asian patient: diagnostic and therapeutic challenges

Giulia Ciccarese,[1] Riccardo Bortone,[2] Alexandre Raphael Meduri,[2] Benedetta Tirone,[2] Francesco Drago,[3] Caterina Foti,[2] Gloria Hoxhallari,[4] Domenico Bonamonte,[2] Marco Bellino[2]

[1]Unit of Dermatology, Department of Medical and Surgical Sciences, University of Foggia, Italy; [2]Section of Dermatology and Venereology, Department of Precision and Regenerative Medicine and Ionian Area, University of Bari "Aldo Moro", Italy; [3]Casa di Cura Villa Montallegro, Genoa, Italy; [4]Department of Dermatology and Venereology, Mother Teresa University Hospital Center, Tirana, Albania

Correspondence: Giulia Ciccarese, Unit of Dermatology, Department of Medical and Surgical Sciences, University of Foggia, Viale Pinto, 1, 71122 Foggia, Italy. Tel.: +393208111421. E-mail: giulia.ciccarese@unifg.it

Key words: Morbihan disease; Asian patient; facial edema; rosacea complication.

Contributions: Marco Bellino, Riccardo Bortone, Alexandre Raphael Meduri, Gloria Hoxhallari, Benedetta Tirone: conception and acquisition of data; Giulia Ciccarese, Domenico Bonamonte, Caterina Foti: analysis and interpretation of data; Giulia Ciccarese, Riccardo Bortone, Francesco Drago: drafting the article and revising it critically for important intellectual content. All authors have read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Conflict of interest: the authors have no conflict of interest to declare.

Ethics approval and consent to participate: not required.

Consent for publication: the patient described in this manuscript has given written informed consent to participate in the study and to the use of his de-identified, anonymized, aggregated data and case details (including photographs) for publication.

Availability of data and materials: the data supporting this study's findings are available from the corresponding author upon reasonable request.

Dear Editor,

Morbihan disease (MD) is a rare condition affecting mainly middle-aged Caucasian adults, characterized by persistent swelling of the upper face. First described in 1956, it was named by Gorin *et al.* in 1991 after a French patient from the Morbihan region.[1,2] The etiology remains unclear and is likely multifactorial, involving lymphatic drainage impairment and chronic vascular inflammation, supporting its association with rosacea.[2-4] Histological findings parallel those of rosacea, characterized by perivascular and inflammatory infiltrates, dermal edema, and sebaceous gland hyperplasia.[2-5] Diagnosis relies on clinical features and ruling out other conditions that look similar.[1-4] MD is notoriously resistant to many treatments, including corticosteroids and antihistamines, and there is no universally accepted effective therapy.[4-6] We report a case of Morbihan disease in a non-Caucasian patient successfully treated with oral isotretinoin, highlighting the diagnostic and therapeutic challenges and contributing to the ongoing debate on its relationship with rosacea.

A 56-year-old previously healthy Mauritian man presented with a two-year history of progressive facial swelling, preceded by erythema that had begun a decade earlier. He denied pain or pruritus, and previous corticosteroid therapy had been ineffective. Dermatological examination revealed persistent erythematous, non-pitting edema involving the cheeks, nose, and right lower eyelid, resulting in incomplete opening of the right eye. Associated findings included erythema, telangiectasias, and papulopustular lesions (Figure 1A). Laboratory and instrumental investigations were within normal limits or negative. Histopathological examination was not performed due to the patient's refusal. A diagnosis of Morbihan disease was established.

Oral isotretinoin was initiated at 20 mg daily. After four months, marked improvement was observed, with residual edema limited to the lower right eyelid. The dose was tapered (20 mg every other day), and topical tacrolimus 0.1% ointment was added three times weekly.

After an additional four months, complete resolution was achieved (Figure 1B); only a mild residual erythema of the cheeks was observed on dermoscopy (Figure 1C). The patient remains in follow-up (18 months) and provided informed consent for publication.

Due to its rarity and clinical overlap with other dermatoses, MD remains a diagnostic challenge. Differential diagnoses include inflammatory, infectious, iatrogenic, neoplastic, and hereditary causes,[1-4] such as Melkersson-Rosenthal syndrome, dermatomyositis, sarcoidosis, erysipelas, cutaneous lymphomas, and thyroid disease.

Melkersson-Rosenthal syndrome, a rare disorder characterized by orofacial edema, facial nerve palsy, and tongue fissuring, was excluded based on persistent edema rather than recurrence and absence of facial palsy.[1,2,7]

Dermatomyositis, an idiopathic inflammatory disease with cutaneous manifestations associated with muscle involvement and systemic complications, was also excluded due to the absence of muscle symptoms and normal laboratory (anti-nuclear and extractable nuclear antigen antibodies, creatine phosphokinase) and chest X-ray findings.[1,3]

Cutaneous sarcoidosis, a multisystem granulomatous disease requiring histological confirmation, was ruled out considering the absence of respiratory symptoms and normal angiotensin-converting enzyme levels and chest X-ray.[1]

Cutaneous lymphoma was considered unlikely given the lack of systemic involvement and complete therapeutic response.[8] Thyroid disease was excluded based on normal laboratory results.[1]

Infectious causes of periorbital edema, such as Epstein-Barr virus infection (Hoagland's sign)[9] and bacterial periorbital cellulitis,[1] were unlikely given the non-acute presentation and absence of fever.[1]

MD should be distinguished from lymphedema. Secondary lymphedema is most commonly caused by filariasis worldwide and by malignancy in developed countries.[1-4] In our patient, filaria was unlikely given his long-term residence in Italy, and cutaneous metastasis was considered unlikely given the gradual, bilateral onset, absence of systemic involvement, and lack of prior oncologic surgery.

This case is notable for the patient's ethnicity and the excellent response to first-line therapy, which allowed treatment to be maintained unchanged over time. Although histopathology was not performed in our patient, the clinical presentation and favorable response to oral isotretinoin supported the diagnosis.

Most studies on MD involve Caucasian patients, suggesting possible under-recognition in darker phototypes due to the difficulty in detecting erythema and differences in UV susceptibility.[5] Similarly, rosacea is less frequently reported in darker skin, likely leading to delayed diagnosis and increased morbidity.[10]

Current evidence suggests that MD may represent a variant or complication of rosacea; however, this remains debated, as it can occur in the absence of typical rosacea features. Unlike rosacea, where small intestinal bacterial overgrowth and *Helicobacter pylori* infection are more prevalent,[10] these gastrointestinal conditions have not been investigated in MD.[1-6]

Based on previous literature reviews,[2,3,6] tetracyclines and isotretinoin emerged as the most effective oral therapies. Topical agents such as tacrolimus and ivermectin may be considered when systemic therapies are not feasible. Overall, data from the literature[5,6] and the present case report showed that clinical features and treatment outcomes in non-Caucasian patients with MD are comparable to those reported in Western populations.

In conclusion, MD can lead to significant functional and aesthetic impairment. It remains a diagnostic and therapeutic challenge, and greater clinical awareness across all phototypes is essential to avoid misdiagnosis and inappropriate management.

References

1. Veraldi S, Persico MC, Francia C. Morbihan syndrome. Indian Dermatol Online J 2013;4:122-4.
2. Mayur O, Martinez R, McNichol MC, McGee JS. Clinical and histological features and treatment outcomes of patients with Morbihan disease: a systematic review. Arch Dermatol Res 2023;315:2427-9.
3. van der Linden MM, Arents BW, van Zuuren EJ. Diagnosis and treatment of Morbihan's disease: a practical approach based on review of the literature. J Clin Aesthet Dermatol 2023;16:22-30.
4. Ramirez-Bellver JL, Pérez-González YC, Chen KR et al. Clinicopathological and Immunohistochemical Study of 14 Cases of Morbihan Disease: An Insight Into Its Pathogenesis. Am J Dermatopathol 2019;41:701-10.
5. Park J, Kim MB, Choi HY, Yang SW. Novel prognostic factors and combination therapy outcomes in Morbihan disease: insights from an Asian population. BMC Ophthalmol 2024;24:496.
6. Yilmaz O, Bose R, Colantonio S. Low-dose isotretinoin for the treatment of Morbihan's disease in a 69-year-old Asian male: A case report. SAGE Open Med Case Rep 2025;13:2050313X251320214.
7. Dhawan SR, Saini AG, Singhi PD. Management Strategies of Melkersson-Rosenthal Syndrome: A Review. Int J Gen Med 2020;13:61-5.
8. Oh Y, Stoll JR, Moskowitz A, et al. Primary cutaneous T-cell lymphomas other than mycosis fungoides and Sézary syndrome. Part II: Prognosis and management. J Am Acad Dermatol 2021;85:1093-106.
9. Ciccarese G, Trave I, Herzum A, et al. Dermatological manifestations of Epstein-Barr virus systemic infection: a case report and literature review. Int J Dermatol 2020;59:1202-9.
10. Drago F, Ciccarese G, Herzum A, et al. Rosacea and alcohol intake. J Am Acad Dermatol 2018;1:25.

Figure 1. A) Erythematous, non-pitting edema affecting the cheeks, the nose and the right lower eyelid, preventing the patient from fully opening the right eye; erythema, telangiectasia, and a few papulopustular lesions were observed on the nose and cheeks; **B**) complete recovery after eight months from the beginning of the treatment with resolution of the erythema, papules, pustules and edema; **C**) dermoscopy (10x magnification, DL5 dermoscope [DermLite, Aliso Viejo, USA]) of the cheeks showed only a slight erythema.