

Late cutaneous reaction after gold patch testing with features of Langerhans cell histiocytosis

Gabriela Pita da Veiga¹  | Laura Sainz-Gaspar¹ | Jose M. Suárez-Peñaranda² | Dolores Sánchez-Aguilar¹ | Hugo Vázquez-Veiga¹ | Virginia Fernández-Redondo¹

¹Department of Dermatology, University Hospital Complex, Santiago de Compostela, Spain

²Department of Pathology, University Hospital Complex, Santiago de Compostela, Spain

Correspondence

Dr Gabriela Pita da Veiga, Department of Dermatology, University Hospital Complex, Travesía Choupana s/n, 15706 Santiago de Compostela, Spain.

Email: gabriela.pitadaveiga@hotmail.com

KEYWORDS: case report, gold patch test, gold sodium thiosulfate, Langerhans cell histiocytosis, metals

Langerhans cell (LC) histiocytosis (LCH) is the preferred term for a spectrum of diseases that can involve different organs, including the skin. LC hyperplasia is a different entity, and may be histopathologically indistinguishable from LCH. Close clinical and pathological observation is mandatory to avoid misinterpretation of the lesions and overdiagnosis of LCH.

CASE REPORT

We present the case of a 57-year-old woman with lesions on both arms (Figure 1A) diagnosed as cutaneous, biopsy-proven LCH 1 year previously. Routine analysis and the imaging studies (skeletal survey, cerebral magnetic resonance imaging, and abdominal ultrasonography) did not show abnormal findings. She did not have a history of atopy or metal intolerance. Because our patient reported itching while wearing dark-coloured clothes, patch testing was performed according to ICDRG criteria with the Spanish Contact Dermatitis Research Group baseline series by the use of TRUE Test, and with supplementary allergens supplied by AllergEaze (Calgary, Canada), as well as with a textile series (Chemotechnique Diagnostics, Vellinge, Sweden). The allergens were prepared in Curatest chambers (Lohmann & Rauscher, Neuwied, Germany), applied to the patient's back, and fixed with Microspore tape.

Patch testing yielded negative reactions to all of the allergens read on day (D) 2, D4, and D7. However, 5 months later, the patient showed an erythematous rectangular plaque coinciding with the application site of gold sodium thiosulfate (TRUE Test; 75 µg/cm²) (Figure 1B). Because of suspicion of a lymphomatoid reaction, a biopsy was performed. Histology (Figure 2) showed superficial dermal nodules of lymphocytes, eosinophils, and large amounts of LCs, which stained with antibodies against S-100 protein, CD1a, and langerin (CD207). The lesion remained unchanged during a follow-up of 15 months.

DISCUSSION

Gold patch testing may be associated with delayed and persistent reactions that might be missed on standard testing or that are difficult to distinguish from active sensitization.¹ Furthermore, different

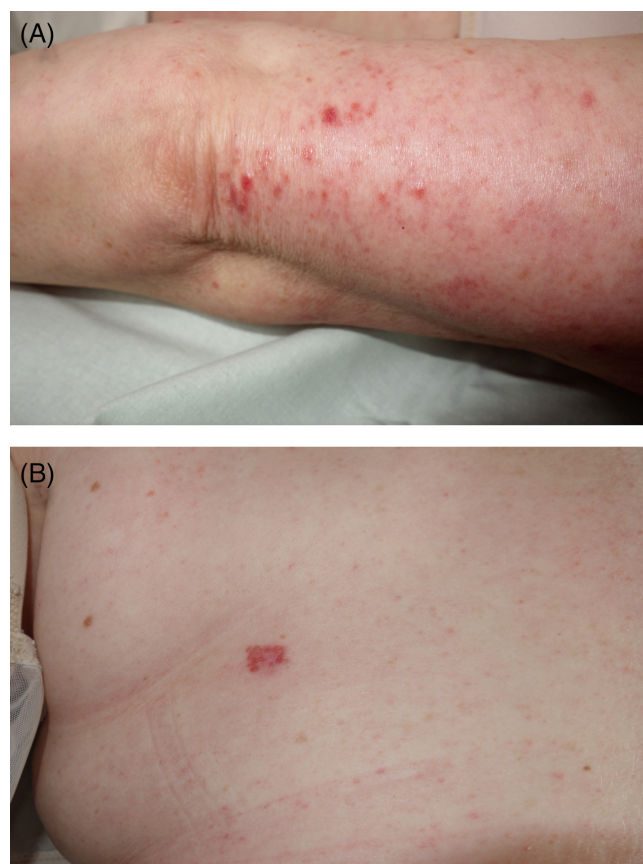


FIGURE 1 Cutaneous involvement of Langerhans cell histiocytosis, showing erythematous papules on the outer side of the upper extremity (A). Five months after the patch testing was performed, the patient showed an erythematous plaque coinciding with the site of the gold sodium thiosulfate patch test (B)

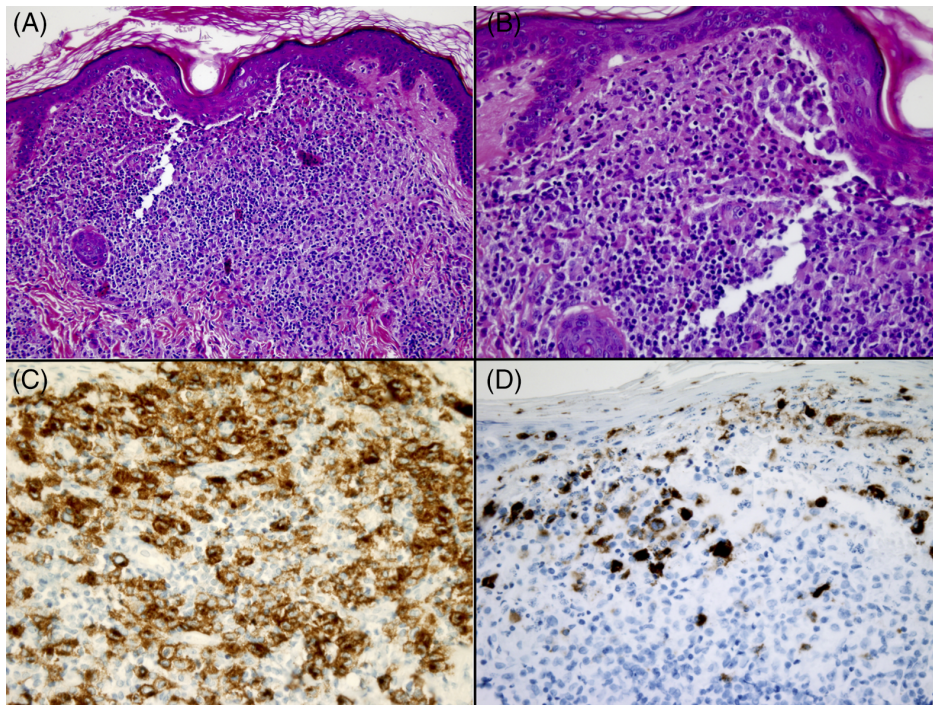


FIGURE 2 Histology showed superficial dermal nodules of lymphocytes, eosinophils and large amounts of Langerhans cells (A, B), which stained with antibodies against CD1a (C) and Langerin (D)

manifestations of allergic contact dermatitis caused by gold have been described.² Our case is highly unusual, as it shows features of LCH, thus creating difficulties in its interpretation. The exact coincidence of the lesion with the site of the patch test makes it difficult to believe that it is a neoplastic lesion, and an isomorphic response has not previously been reported in LCH.³

Alternatively, a hyperplastic response triggered by gold sodium thiosulfate could be considered, but differential diagnosis with a true LCH lesion would be extremely difficult in our patient. Several previous reports have shown LC hyperplasias in different clinical contexts, mainly haematological malignances⁴ or scabies,⁵ which are conditions that were not present in our case. Immunostaining for p53, cyclin D1 and CD31 has also been reported as a supplemental investigation for differential diagnosis.^{6,7} Our patient showed diffuse positivity for CD31 and cyclin D1 in the dermal component, and focal positivity for p53 in both biopsies. These results support the neoplastic nature of both lesions. Recently, *BRAF* V600E mutations have been shown in LCH, but they were present in no more than 64% of the patients.⁸ In our patient, it was absent both in the biopsy of one of the LCH lesions and in that from the patch test site.

In conclusion, our patient presents a previously unreported late reaction at the site of gold sodium thiosulfate patch testing. The true nature of this lesion, hyperplastic or neoplastic, cannot be definitely elucidated.

CONFLICTS OF INTEREST

Authors declare no conflict of interest.

ORCID

Gabriela Pita da Veiga  <https://orcid.org/0000-0002-6683-8370>

REFERENCES

1. Bruze M, Hedman H, Björkner B, Möller H. The development and course of test reactions to gold sodium thiosulfate. *Contact Dermatitis*. 1995;33:386-391.
2. Chen JK, Lampel HP. Gold contact allergy: clues and controversies. *Dermatitis*. 2015;26:69-77.
3. Sagi L, Trau H. The Koebner phenomenon. *Clin Dermatol*. 2011;29: 231-236.
4. Christie LJ, Evans AT, Bray SE, et al. Lesions resembling Langerhans cell histiocytosis in association with other lymphoproliferative disorders: a reactive or neoplastic phenomenon? *Hum Pathol*. 2006;37: 32-39.
5. Bhattacharjee P, Glusac EJ. Langerhans cell hyperplasia in scabies: a mimic of Langerhans cell histiocytosis. *J Cutan Pathol*. 2007;34: 716-720.
6. Grace SA, Sutton AM, Ambrecht ES, Vidal CI, Rosman IS, Hurley MY. p53 is a helpful marker in distinguishing Langerhans cell histiocytosis from Langerhans cell hyperplasia. *Am J Dermatopathol*. 2017;39:726-730.
7. Shanmugam V, Craig JW, Hornick JL. Cyclin D1 is expressed in neoplastic cells of Langerhans cell histiocytosis but not reactive Langerhans cell proliferations. *Am J Surg Pathol*. 2017;41:1390-1396.
8. Harmon CM, Brown N. Langerhans cell histiocytosis. A clinicopathologic review and molecular pathogenetic update. *Arch Pathol Lab Med*. 2015;139:1211-1214.

How to cite this article: Pita da Veiga G, Sainz-Gaspar L, Suárez-Peñaranda JM, Sánchez-Aguilar D, Vázquez-Veiga H, Fernández-Redondo V. Late cutaneous reaction after gold patch testing with features of Langerhans cell histiocytosis. *Contact Dermatitis*. 2019;81:291-292. <https://doi.org/10.1111/cod.13281>