syndrome are sometimes associated with acrokeratosis paraneoplastica (Bazex syndrome) [8]. Although nail dystrophy is common in Bazex syndrome, our patient showed no eruption on other commonly affected parts, such as the soles of the feet, nose, and earlobes, and the histological feature of the skin lesion was a typical pattern of psoriasis. Moreover, the histological analysis of BC indicated papillo-tubular carcinoma, which has never been reported to be associated with Bazex syndrome [8]. On the basis of these findings, we excluded a diagnosis of Bazex syndrome. The biological reason for the association between psoriasis and BC is unclear [1-3]. Some reports have suggested significant roles of interleukin (IL)-17, IL-23, and tumour necrosis factor (TNF)-α in tumour immunity in patients with BC [9, 10]. Moreover, these cytokines are targeted for treating psoriasis [4-6]. We speculate that these increased cytokines in patients with BC may play a role in the development of psoriasiform eruptions. A limitation of this report is that the change in IL-17, IL-23, and TNF-α levels was not examined, however, blood levels of white blood cells, neutrophils, and C-reactive protein decreased to within normal range after resection of BC. The resection of BC and disappearance of psoriasiform eruptions may have had an effect on this change. The expression site of skin eruptions was characteristic in our case. Our patient had skin eruptions mainly on her scalp and nails with severe pruritus. We therefore describe a case with distinctive expression of psoriasiform eruptions possibly associated with BC, which has not been previously reported.

In conclusion, we diagnosed and curatively treated a patient with BC at an early stage, who had no symptoms, except for psoriasiform eruptions. Although further studies are needed to clarify the pathological mechanism of the association between psoriasiform eruptions and BC, our report highlights the possibility of psoriasiform eruptions as a paraneoplastic syndrome of BC.


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Progressive nodular histiocytosis improved by methotrexate

Non-Langerhans cell histiocytes (NLHs) represent a heterogeneous group of uncommon disorders [1]. Their classification, based on clinical, histological, immuno-histochemical, and evolving features, is difficult [2]. Moreover, both mixed and transitional forms between the different entities have been described. The rarity of cases and existence of different spontaneously-regressive entities account for the lack of standardized treatments. The treatment is often disappointing and therapeutic successes, as in our case, should be reported to improve patient support.

A 78-year-old woman developed an eruption of brown papules on her back suggesting xanthoma disseminatum (XD) (figure 1A). The lesions spread to the neck, trunk, limbs, palms, and scalp, and were accentuated in flexural areas. Histopathological examination of a skin biopsy revealed a dermal infiltrate with histiocytic and epithelioid cells forming nodules, consistent with an NLH. Further work-up (CT-scan, TEP-scan, and trephine biopsy) did not reveal extracutaneous involvement. Despite topical treatment with betamethasone, a new flare appeared. The lesions progressed to yellow-brown papules and were associated with lymphadenopathic appearance; they became widespread, voluminous, nodular, and pruritic, resulting in a leonine aspect on the face (figure 1B). Some lesions on the back became necrotic (figure 1C). This new appearance was typical of progressive nodular histiocytosis (PNH). Histology of a papular lesion showed diffuse dermal infiltration with histiocytes, some of which were foamy (figure 1D). The cell nuclei were uniform. These cells were CD68-positive but CD1a and S100 protein-negative, consistent with NLH. In addition, numerous eosinophils,
Disappearance of lesions without new flare-up after treatment. D, E, F) Disappearance of lesions without new flare-up after treatment with methotrexate. G) Histology of a small papular lesion showing a diffuse infiltrate (hematoxylin & eosin [H&E] and safran staining; original magnification: ×100). The dermis contains abundant histiocytes, some with a foamy cytoplasm. The cell nuclei are uniformly shaped. H, I) In addition, there are few multinucleated giant cells and scattered inflammatory cells, including lymphocytes, plasma cells, and numerous eosinophils (H&E; ×400). Immunohistology (not shown) was positive for CD68 but negative for CD1a and S100.

Some multinucleated giant cells, scattered lymphocytes, and plasma cells were seen (figure 1H, I). The course was favourable after oral administration of 70 mg/d prednisone, but intercurrent infectious pneumonia led to treatment discontinuation after two months. A relapse of the skin lesions associated with pruritus was observed after two weeks off-treatment. Oral methotrexate (15 mg once/week) led to dramatic reduction of pruritus associated with the resolution of the lesions. After six months, the patient presented an almost complete clinical response without relapse and remained in remission for 18 months after starting methotrexate. However, methotrexate was discontinued because of urinary tract infection that necessitated hospital admission. After discharge, a new flare of PNH lesions occurred on the face, lower neckline, and arms. Methotrexate treatment was reintroduced as before, and remission was once again obtained within three months. The patient had no relapse within six months (figure 1D-F).

NLHs are characterized by the proliferation of non-Langerhans dendritic cells (e.g., dermal dendrocytes or intermediate cells). They include different entities belonging to the same spectrum [1], such as Erdheim-Chester disease, PNH, Rosai-Dorfman disease (RDD), and XD. These disorders can have overlapping features [3, 4]. In our patient, the lesions progressively evolved from XD to PNH. PNH is a rare type of NLH, in which two types of lesions can be observed: yellow-brown or yellow-pink papules, along with widespread deep nodules with occasional prominent facial involvement [2]. Usually, these lesions do not regress spontaneously [2, 5]. Additionally, our patient suffered from severe pruritus, which has not been previously described in PNH.

No consensus treatment for NLHs exists yet, and treatment outcomes are often disappointing. In our patient, corticosteroids showed incomplete efficacy, but the disease was improved by low-dose methotrexate. Methotrexate has been used in some cases of systemic RDD [6] and reticulo-histiocytoma [7]. Randrianosolo et al. also used methotrexate in PNH with temporary (three-month) efficacy [8]. As far as we know, no effective treatment has been reported for PNH, other than surgical removal of nodules with high functional or aesthetic impact [5, 9]. Prednisolone, vincristine, and cyclophosphamide have proven unsuccessful [10]. These lesions can be disfiguring (leonine facies) and impact significantly on quality of life. In our patient, methotrexate proved effective on two occasions, therefore it should be considered for the treatment of NLHs, especially in PNH.