

Hirudotherapy – a rare cause of pseudolymphoma

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Cutaneous pseudolymphoma (C-PSL) represents a heterogeneous group of cutaneous reactions characterized by polyclonal T and/or B cell proliferation [1, 2]. The disease can be either idiopathic or triggered by various stimuli including infections, primarily *Borrelia burgdorferi* and drugs [3, 4]. Several lines of evidence indicate minor traumas such as tattoos, acupuncture, insect bites and vaccination as C-PSL inducers [3, 4]. Herein, we present a rare case of a C-PSL provoked by hirudotherapy.

A 38-year-old woman was admitted to the Department of Dermatology, Venereology and Allergology, Wrocław Medical University, for diagnosis and treatment of skin lesions that appeared 7 months before hospitalization. On admission, physical examination revealed 10 well-defined, firm and pruritic nodules, red in color, ranging from 6 to 10 mm in diameter in her pubic area (Figures 1 A, B). The skin changes appeared soon after the patient underwent hirudotherapy that had been recommended by her friend as a successful treatment for uterine myoma. The pruritic cutaneous lesions appeared after the first procedure, however the patient decided to apply the second therapy after 3 weeks. Each time 5 leeches were used. For the apparent skin lesions the patient was given oral antihistamines and moderate potent topical glucocorticosteroids with no improvement. The patient did not take any other medicines, except for ulipristal acetate 5 mg/day, orally, administered by a gynecologist for the uterine myoma.

Laboratory tests (morphology, urinalysis, lipids, liver function, renal function and inflammation markers) were within normal ranges. Histological examination of the skin biopsy revealed irregular acanthosis and mixed-cell infiltration, composed of lymphocytes, histiocytes, plasma cells and eosinophils in the dermis. An evident exocytosis with focal spongiosis and intraepidermal inflammatory infiltration were observed. In the immunohistochemical study, the lymphocyte population consisted of a mixed population of T and B lymphocytes with

T cell predomination. The T lymphoid cells were positive for CD3, CD43, CD45RO, CD4, CD8, CD30, CD20, CD79a, CD138. The numerous epithelioid histiocytes were CD68 positive. The proliferative rate measured by Ki-67 expression was low (labeling index = 7%). Based on the histological, immunohistochemical results and clinical observations the diagnosis of C-PSL was made.

Initially, the skin changes were treated with 100 mg hydrocortisone intralesionally and topical glucocorticosteroid ointment (clobetasol propionate) applied twice a day. Cryotherapy was used in the case of one nodule, however the results were unsatisfactory as the nodule became discolored. After 4 weeks of topical treatment, nodules became smaller and less red but remained firm. We modified the previous therapy by adding methylprednisolone acetate intramuscularly 40 mg every 4 weeks. We chose intramuscular instead of intralesional drug administration because the patient had a bad experience with previous mesotherapies and preferred systemic treatment (one injection) instead of a series of injections. The patient continued treatment for 20 weeks with a slow regression of skin lesions.

Cutaneous pseudolymphomas are not a rare dermatological problem, however, due to the overlapping histopathological and clinical features they may create a diagnostic and therapeutic challenge even for experienced dermatologists [5, 6]. To diagnose C-PSL, histological and clinical criteria must be fulfilled [5, 6]. Clinical criteria include typical location and morphology of the skin changes as well as characteristic clinical course of the disease. Typically, skin changes appear on the face, which is the most common localization of the lesions, on the chest and upper extremities. They are usually single and localized nodules or plaques, red to purple in color and tend to be self-regressing or disappear when the causative factor is removed [3]. Histological features of C-PSL include polyclonal lymphocyte infiltration in the upper layers of the skin. According to the predominant

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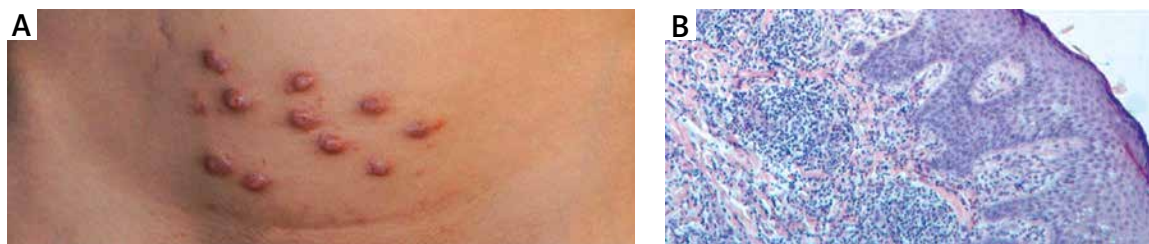


Figure 1. A – A 38-year-old female with skin changes in the pubic area after hirudotherapy. **B** – A significant inflammatory infiltration in the dermis composed mostly of lymphocytes and eosinophils. An evident epidermotropism with spongiosis was observed in some parts of the infiltration (hematoxylin and eosin, magnification 100×)

lymphocyte type in the infiltrate, several types of cutaneous pseudolymphomas have been distinguished: C-PSL with predominant T-cell (T-PSL), B-cell (B-PSL) and mixed cell infiltrations. C-PSLs with mixed cell infiltration, as in our case, usually occur after acupuncture, vaccination, tattoos and in *Borrelia burgdorferi* infections. T-PSL infiltration was reported as the effect of persistent scabies, chronic contact dermatitis, arthropod bites, drug reaction and in idiopathic cases [7]. B-PSLs are often related to drug-induced reactions [8].

Hirudotherapy has been used in the traditional medicine for bloodletting for medical conditions such as chronic venous insufficiency and muscular pain as well as cosmetic reasons. The complications of the procedure include infections, local irritation, scarring, bleeding and anemia. To the best of our knowledge, this is the first case of pseudolymphoma after hirudotherapy reported in Poland, and there have been only three such cases described in the world literature [9–11]. In the published reports medicinal leeches were applied for fibromyalgia, chronic venous insufficiency with varicosis, and, in 1 case, for cosmetic reasons (black circles in the infraorbital regions). In contrast to our case, where we found mixed B and T-cell infiltrate in the skin biopsy, the biopsies taken from the skin lesions in the reported cases revealed predominant B-cell infiltrates in two cases and T-cell infiltrate in 1 case. Topical and intralesional glucocorticosteroids, antibiotics, ultraviolet B phototherapy, cyclosporine A, interferon $\alpha 2\beta$, laser therapy and surgical excision have been reported as a successful therapy of the pseudolymphomas [12]. The reported cases of pseudolymphomas after medical leeches were treated successfully with either topical mometasone furoate or intralesional triamcinolone acetonide injections with complete clearing of the lesions [9–11].

In recent years there has been increased interest in alternative methods of treatment, including hirudotherapy. However, we should always keep in mind undesirable side effects of these procedures such as development of pseudolymphoma.

Conflict of interest

The authors declare no conflict of interest.

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