

CASE REPORT

Multiple nodules on the left cheek represented pseudolymphomatous folliculitis

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Introduction

Pseudolymphomatous folliculitis (PLF) is a rare lesion of which few reported cases exist. The clinical appearance is characterized by solitary or multiple large, firm violaceous nodules that occur mainly on the face, scalp, and upper trunk [1]. Typically, rapid growth is seen over a period of several weeks or months [1], making distinguishing PLF from malignant lymphoma clinically difficult. We treated a case of subcutaneous PLF on the left cheek and herein present the characteristic pathological findings.

Case Report

A 51-year-old male visited our hospital with multiple rapidly enlarging masses in the left cheek that had been developing for 1 month. He had a history of cerebral infarction, and clopidogrel sulfate had been administered to prevent another infarction. He also had diabetes, for which he was under medical treatment. Physical examination revealed three firm, red, dome-shaped subcutaneous nodules on the left cheek, two of which were located between the ear and the jaw, with the remaining one near the nose. The three nodules measured 7 × 10, 6 × 5,

Key Clinical Message

Pseudolymphomatous folliculitis (PLF) is a rare lesion. Sometimes, the clinical appearance is characterized by multiple large, firm violaceous nodules. In cases with multiple lesions, such biopsy should be performed on one lesion, and once PLF is determined, monitoring for the remained tumor is considered to be the best treatment.

Keywords

Multiple, pseudolymphomatous folliculitis.

and 5 × 5 mm, respectively. None of the masses were adhered to the connective tissue (Fig. 1A). Based on our suspicion of inflamed epidermal cysts, surgical excision of the two lesions between the ear and the jaw was performed under local anesthesia 1 month after the initial examination. With consideration of the influence of inflammation, those two tumors were removed along with fat in the immediate area surrounding the tumors. We did not remove all three tumors at the same time because of the risk of hemorrhage due to the anticoagulant medication. After confirming that postsurgical bleeding was controlled, the remaining tumor, located near the nose, was removed 12 days after the first operation, under local anesthesia (Fig. 1B).

Pathological findings of the tumors excised at the first operation showed a nodular dense lymphoid cell infiltration from the dermis to the subcutaneous tissue. The cells had been enlarged and the hair follicles had been destroyed by infiltration of the dense, diffuse, and small well-differentiated lymphocytes (Fig. 2A–C). Furthermore, immunohistochemical examination revealed that a large number of lymphocytic cells were positive for CD3, S-100, and CD1a (Fig. 2D, F and G), while few were positive for CD20 and few were positive for PD-1 (Fig. 2E



Figure 1. Preoperative view of left cheek (A). Preoperative view of the tumor near the nose, taken 12 days after the first excisional biopsy for the other two tumors (as indicated by the arrow), showing slight regression after the first visit (B).

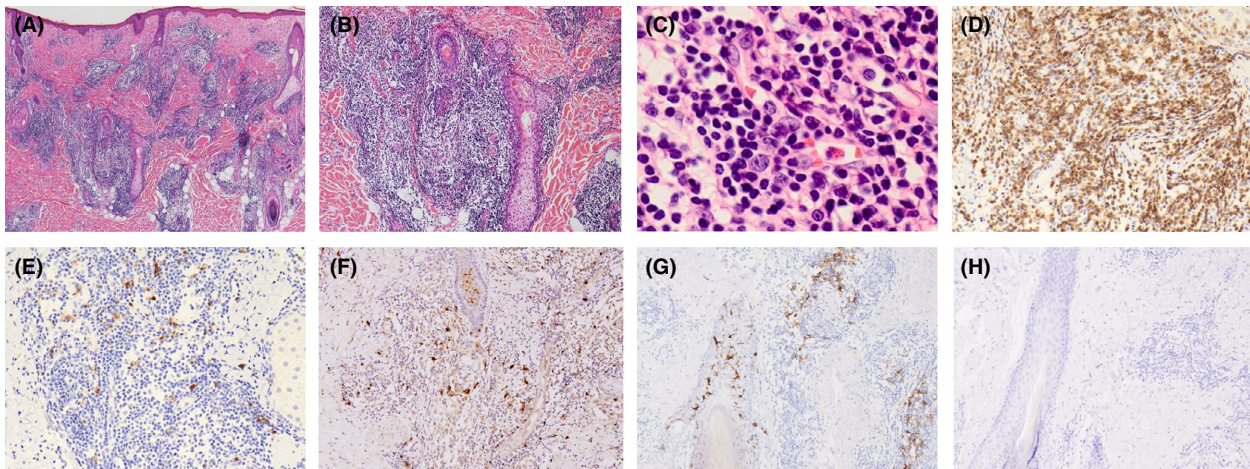


Figure 2. Enlarged and distorted hair follicles are surrounded by a dense diffuse infiltration of cells (A, B). The infiltrating cells were mainly composed of small lymphocytes with some blast cell-like large cells (C). Immunostaining revealed sheets of CD3+ T lymphocytes (D) and scattered CD20+ B lymphocytes (E). Furthermore, a large number of lymphocytic cells were positive for S-100 and CD1a (Fig. 2F and G), whereas few lymphocytic cells were positive for PD-1 (Fig. 2H). (magnification: A, $\times 2$; B, $\times 10$; C, $\times 40$; D-H, $\times 10$).

and H) [2]. Based on these findings, we made a diagnosis of PLF.

However, in a pathological examination of the excised whole tumor at the second operation, mild lymphocyte infiltration around the hair follicles was found, which was not considered to be a PLF-specific finding (Fig. 3). Thus, from a pathological point of view, the lesion was considered to be nonspecific inflammation. Taking all findings into account, the patient was diagnosed as having mild or regressed PLF. Six months after the second operation, healing progressed and no recurrence was noted (Fig. 4).

Discussion

PLF is characterized by a dense lymphoid infiltration accompanied by hyperplastic hair follicles, and was first

described as a distinct variant of pseudolymphoma in 1986 [3]. Thereafter, only approximately 10 reports on PLF, including 76 cases, have been published; thus, it is considered to be a rare entity. According to those past reports, typical PLF is presented as a solitary, erythematous or violaceous, dome-shaped or flat elevated nodule on the face, especially on the nose, cheek, eyelid, or forehead [4, 5]. In many cases, the eruption is isolated [5], whereas our patient showed a relatively rare pattern of eruption, as the PLF manifested as multiple lesions. PLF occurs in a wide age range and both sexes [5]; however, its tendency is yet to be elucidated because of the low number of reports. To the best of our knowledge, no cases of spontaneous regression without any treatment (operation, biopsy, medication, etc.) have been reported.

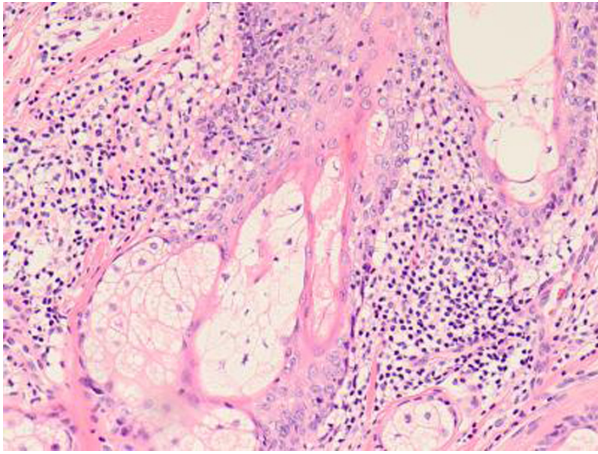


Figure 3. Mild lymphocyte infiltration was found around the hair follicles (magnification: $\times 4$).



Figure 4. No recurrence was noted 6 months after the second surgical procedure.

A variety of differential diagnoses, including epidermal cyst, chronic folliculitis, granuloma, basal cell carcinoma, sarcoidosis, and insect bite reaction must be considered [1, 5], with malignant lymphoma being one of the most important. Thus, careful histological and immunohistochemical examinations are needed for accurate diagnosis of PLF [1]. Arai *et al.* [4] proposed histological criteria for diagnosis, including nodular dense lymphocytic infiltration from the dermis to subcutis, befitting the term pseudolymphoma, in which lymphocytes surround and infiltrate the pilosebaceous unit, and deform the walls. Kazakov *et al.* [5] presented 42 cases of PLF in which hair follicles were enlarged and often distorted, with a dense diffuse lymphoid infiltration composed mainly of small well-differentiated lymphocytes, occupying the whole dermis, though sparing of the epidermis was also observed.

Immunohistologically, PLF can be divided into four groups: predominance of B cells, predominance of B cells with fairly numerous T cells, predominance of T cells with fairly numerous B cells, and predominance of T cells [4]. Recently, Goyal *et al.* [2] reported that PD-1+ T cells and CD1a are useful for differentiation of PLF. In our case, distorted hair follicles and diffuse lymphocyte infiltration with no dysplasia were found (Fig. 2A–C), as well as sheets of CD3+ T lymphocytes and scattered CD20+ B lymphocytes (Fig. 2D and E). Furthermore, a large number of lymphocytic cells were positive for S-100 and CD1a (Fig. 2F and G), whereas few lymphocytic cells were positive for PD-1 (Fig. 2H). Based on these findings, we made a diagnosis of PLF. In contrast to lymphomas, lymphocytes in PLF show no bias with regard to their κ -/ λ -chain-positive B-cell ratio or CD4+/8+ T-cell ratio. In addition, the results of polymerase chain reaction assays for clonal T-cell receptor and immunoglobulin heavy chain gene rearrangements typically reveal negative findings in PLF cases [6]. Since we already had a diagnosis, we did not perform additional immunohistological examination. On the other hand, in the specimen obtained in the second operation, we found only mild lymphocyte infiltration around the hair follicles and common findings of inflammation; therefore, a diagnosis of PLF could not be made from that evidence alone.

The first choice of treatment for PLF is excisional biopsy, with the use of corticosteroid injections recommended if the excision is not complete [1]. In cases with multiple lesions, such a biopsy should be performed on one lesion, and once PLF is determined, monitoring of any remaining tumors is considered to be the best treatment.

There are a few reports of PLF nodules regressing spontaneously within 6 months after excisional biopsy [7, 8]. In cases of PLF diagnosed by partial biopsy, it is recommended to follow-up for half a year, and wait for spontaneous improvement.

Conclusion

Careful examination of patients with PLF is required because distinguishing PLF from malignant lymphoma is difficult. In such cases, an excisional biopsy is the most useful method for making an accurate diagnosis. In consideration of the difficulty of diagnosis, an excisional biopsy is valuable; however, once PLF is diagnosed in one of the multiple tumors by excisional biopsy, such a biopsy is thought to be unnecessary for the remaining tumors.

Conflicts of Interest

There are no conflicts of interest to report.

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