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Perifolliculitis Capitis Abscedens et Suffodiens in a Caucasian: Diagnostic and Therapeutic Challenge

Liborija Lugović Mihić[1], Davor Tomas[2], Mirna Šitum[1], Iva Krolo[1], Klaudija Šebetić[1], Ines Sjerobabski-Masnec[1], Freja Barišić[1]

[1]Department of Dermatology and Venereology; [2]Ljudevit Jurak University Department of Pathology, Sestre milosrdnice University Hospital Center, Zagreb, Croatia

Corresponding author:

Assist. Professor Liborija Lugović Mihić, MD, PhD
Department of Dermatology and Venereology
Sestre milosrdnice University Hospital Center
Vinogradska cesta 29
HR-10000 Zagreb
Croatia
liborija@yahoo.com

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SUMMARY Perifolliculitis capitis abscedens et suffodiens or dissecting cellulitis of the scalp is a rare, chronic destructive folliculitis of the scalp, characterized by painful nodules, purulent drainage, sinus tracts, keloid formation and cicatricial alopecia. The cause of the disease is unknown, but it is similar in many features to hidradenitis suppurativa and acne conglobata. In our case report, the patient's dermatologic appearance included one slightly erythematous, infiltrated alopecic area with draining lesions in the right parietal part of the scalp with a few alopecic areas in other parts of the scalp. The identification of the infectious agent, repeated swabs and KOH examination/or fungal cultures and tissue sampling for histopathologic analysis were necessary to confirm the diagnosis of perifolliculitis capitis abscedens et suffodiens. The patient received systemic antibiotics (azithromycin and amoxicillin-clavulanate) and oral antimycotic therapy (fluconazole), followed by a long period of oral isotretinoin with local skin care, which led to resolution and thus inhibited the evolution to scarring and nodular stage of the disease. Thus, such combined approach could be useful for other patients with these dermatologic problems.

KEY WORDS: perifolliculitis capitis abscedens et suffodiens, dissecting cellulitis, therapy, isotretinoin

INTRODUCTION

Perifolliculitis capitis abscedens et suffodiens or dissecting cellulitis of the scalp is a rare, chronic destructive folliculitis of the scalp, characterized by painful nodules, purulent drainage, sinus tracts, keloid formation and cicatricial alopecia (1-5). The disease was first described by Spitzer in 1903 and was named by Hoffmann in 1908 (6). It occurs almost exclusively in young adult men and may be more common in black men in their second to fourth decade of life. The disease is rarely seen in Caucasians, although a few such patients are reported in the literature (1,2,7).

The cause of perifolliculitis capitis is unknown, but today it is considered as dissecting folliculitis of terminal hair follicles. The disease is similar in many features to hidradenitis suppurativa and acne conglobata (previously all three were described as the follicular occlusion) (1,8,9).

It is considered that the pathogenesis of the disease is based on the accumulation of material in the follicles, which leads to follicle dilatation and then rupture (10). Keratin and bacteria from the ruptured follicles can initiate a neutrophilic and granulomatous

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response. It most likely represents a primary inflammatory process with secondary bacterial infection (usually with *Staphylococcus aureus* or *Staphylococcus epidermidis*) (10). Furthermore, it is believed that perifolliculitis capitis abscedens et suffodiens more closely, albeit imprecisely, describes the recurrent draining scalp cysts termed 'dissecting cellulitis' by others (11). The disease usually starts as folliculitis on the scalp vertex or occiput. It expands into patches of perifollicular pustules, nodules, abscesses and sinuses. Thus, the lesions begin as multiple firm scalp nodules that rapidly develop into fluctuant ridges that eventually discharge purulent material leading to hair loss (2). It leads to persistent inflammation and destruction of the follicles, and the skin may be covered with crusts and scales; pressure on one fluctuant area may result in multiple follicular openings (8,12).

The disease may resemble acne keloid, but acne is usually located on the nape and rarely has pus or abscesses. In differential diagnosis, it may also be similar to pseudopelade of Brocq, inflammatory tinea capitis, folliculotropic mycosis fungoides, central centrifugal scarring alopecia, etc.

Histopathologic features reveal moderately dense, perifollicular lymphocytic inflammation affecting the lower half of the dermis, extending down into the subcutaneous fat, necrosis and scarring. Thereby, in the acute suppurative stage, a polymorphous infiltrate rich in neutrophil leukocytes surrounds and destroys the deepest portion of the hair follicle. In later chronic stages, keratin remnants lead to a granulomatous reaction. Smear of pustular content is not diagnostic, as a wide variety of organisms can be found, but often gram-negative bacteria prevail (7).

Perifolliculitis capitis is difficult to treat, as it often shows remarkable resistance to conservative management with antibiotics, retinoids, incision and drainage, x-ray epilation, or steroid treatment (1,13). Generally, acute flares are best treated with broad-spectrum antibiotics. Systemic corticosteroids may interrupt the inflammatory response and allow for more rapid healing. There are good results in some patients with subsequent treatment with isotretinoin (1 mg/kg/day/12-20 weeks) (2). Topical therapy includes disinfectant solutions such as chinosol. Epilation of involved follicles may reduce inflammation and its spreading. When the inflammatory process resolves, surgical excision of scarred areas is recommended (2).

The disease is chronic and flares are very frequent. Although recurrences of the disease are often seen, they are generally mild to moderate and appear 6 months to 1 year after treatment (1).

CASE REPORT

The patient presented was admitted to our department for analysis and treatment of multilocular alopecic and slightly erythematous infiltrated areas of the scalp. The patient's history revealed that he had noticed erythematous changes of the scalp in the form of 'pimples' 5 months before and had treated them locally with a drug given to him by his physician (he had no medical documentation), without any improvement. Two months before admission, the patient noticed aggravation of his state with pain and hair loss in the before mentioned areas. On admission, the patient's dermatologic appearance included one slightly erythematous, infiltrated alopecic area in the right parietal part of the scalp, around 4 centimeters in size, with a central yellow crust and a few alopecic areas up to 1 cm in size in other parts of the scalp (Fig. 1). Although clinically the changes appeared as alopecic areas, incision and aspiration showed a purulent content that was referred for analysis, indicating abscess-alopecia. Native mycologic findings and culture of the material were sterile. Bacterial analysis of the scalp swab revealed only saprophytic bacteria.

Histopathology of the scalp biopsy specimen yielded the following finding (Fig. 2); histologically, a slightly acanthotic and spongiotic, ortho- and parakeratotic epidermis is seen on the surface of the retrieved material. The dermis contains rich infiltrates of lymphocytes, plasma cells and eosinophils, as well as rare neutrophils, especially around hair follicles. One part of the dermis contains a small group of giant cells of foreign body type and Langhans. A slight fibrous tissue multiplication is noticed around hair follicles. No abscess was found in the received specimen. Histochemical staining (PAS) showed sporadic struc-

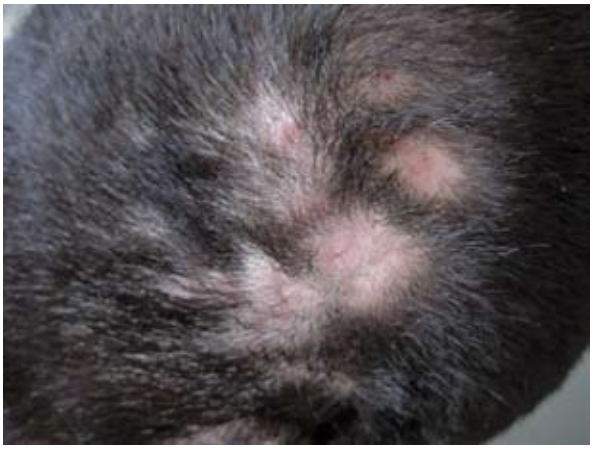

Figure 1. Skin changes with alopecic area in the right parietal part of the scalp.

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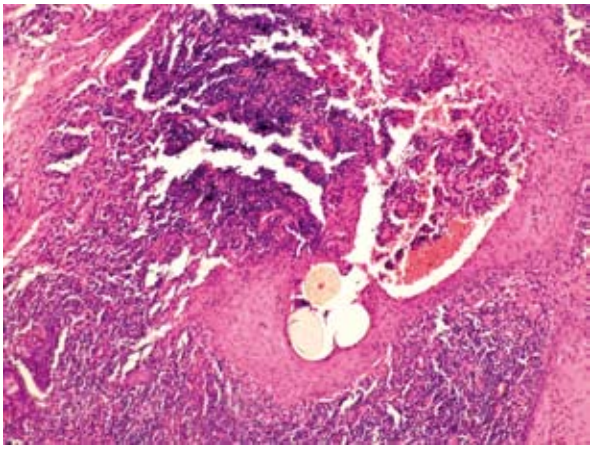

Figure 2. The dermis contains rich infiltrates of lymphocytes, plasma cells and eosinophils, as well as rare neutrophils, especially around hair follicles. A slight fibrous tissue multiplication is noticed around hair follicles. No abscess was found in the specimen. HE; x40.

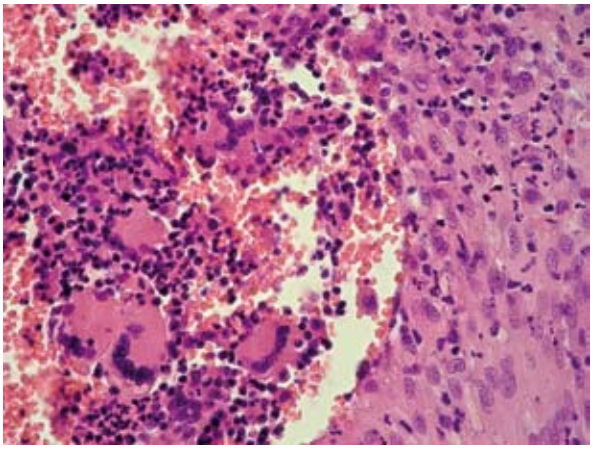

Figure 3. Repeated biopsy showed a small cluster of multinuclear giant cells of foreign body and Langrehans cells in the dermis. HE; x400.

tures that could be referred to as fungal spores, and comprehensive microbiologic (mycologic) evaluation was recommended. Repeat mycologic analysis of the skin scalp was native negative, while subsequently retrieved culture verified *Candida* spp.

Complete blood count (CBC) and differential blood count, erythrocyte sedimentation rate (ESR), serum biochemistry profile (including liver function tests, cholesterol and triglyceride levels), urine analysis, serum albumin, and electrolytes were mostly within the normal limits, with slightly increased ALT and lipid levels.

Other findings were also normal: blood glucose, iron, electrolytes, proteins, AST-O, ASTA, ANA, VDRL, nasopharyngeal swab. Other analyses also yielded normal findings (stool test for bacteria, mycology, parasitology, ECG, chest X-ray, urologist examination, ENT examination, and internal medicine examination).

Based on histology findings, a histopathologist was consulted and interpreted the results as unspecific, with some elements of folliculitis and mycotic infection, and suggested repeat scalp biopsy. During his stay at our department, the patient received systemic antibiotics (azithromycin) for five days, and later amoxicillin-clavulanate for ten days. We also included oral antimycotic therapy (fluconazole capsule 50 mg *per* day for 3 weeks). Locally, we used disinfectant spray and combined local compounds (antibiotic, antimycotic, corticosteroid) for a period of 10 days, followed by local antimycotic clotrimazole cream twice a day. Repeated biopsy was referred for histopathology and microbiology and mycologic tests. Biopsy result (Fig. 3): histology shows orthokeratotic, normal epidermis on the surface of the material. Two follicle orifices contain small keratin taps. The dermis shows perifollicular signs of mild fibrosis and thick infiltrates of mononuclears, rare polymorphonuclears, and some eosinophils. One location in the dermis contains a small cluster of multinuclear giant cells of foreign body and Langerhans type. Histology and histochemistry results (PAS) correspond to the initial diagnosis of perifolliculitis abscedens et suffodiens.

Repeated bacterial tissue culture for aerobes revealed *Staphylococcus aureus* sensitive to cloxacillin, cephalexin, clindamycin, gentamicin, and clotrimoxazole. Mycologic analysis of the scalp skin: native and culture were negative.

Repeated routine laboratory tests (ESR, CBC, electrolytes, liver and kidney function tests, blood lipids) were normal, except for slightly increased cholesterol level (5,9). Nasopharyngeal swab results were normal. Consultant ophthalmologist examined the patient and found no contraindications for retinoid therapy.

According to histopathology results that confirmed the initial diagnosis and skin tissue analysis for aerobes that revealed *Staphylococcus aureus*, oral antibiotic therapy according to the antibiotic sensitivity report was administered (cloxacillin caps 4x500 mg for 10 days). Following therapy with systemic antibiotic administered during his hospital stay, the patient presented for follow up examination and isotretinoin was introduced in therapy. He started using isotretinoin 80 mg *per* day, then tapered off to 60 mg *per* day after one month. The patient noticed dryness of

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his lips and skin associated with his therapy, while scalp lesions stabilized without any further changes. New laboratory test results were normal, except for blood lipids that were slightly increased. The patient received isotretinoin tablets for 3.5 months (total cumulative dose of 50 mg/kg body weight). Because of drug side effects, he also used emollient compounds for his waist skin and lips, as well as photoprotection.

Two years later, the patient was free from disease relapse and had normal hair growth.

DISCUSSION

Perifolliculitis capitis often poses diagnostic problems in dermatology and shows considerable resistance to conservative management. Bacteriologic analysis and tissue culture from skin lesions are usually done; however, results of cultures from unopened lesions tend to be sterile, whereas those from draining lesions yield numerous pathogenic and nonpathogenic organisms (1). Considering the high frequency of sterile cultures, especially those obtained from aspirates of previously unopened lesions, it is believed that bacterial infection is not of central importance in the pathogenesis of the disease, but rather a secondary event. Generally, the culture should guide us to the appropriate choice of antibiotics to manage secondary infections, as in our case. Antibiotics are mostly associated with local care and drainage, and have been the cornerstone of treatment of early disease (13,14).

Very few cases of perifolliculitis capitis in Caucasians have been reported in the literature, and we present one of them. We emphasize that clinical features of perifolliculitis capitis and the corresponding histologic picture may vary from case to case, leading to diagnostic problems. There are also problems related to therapeutic modalities applied and described in the literature, which are limited and often disappointing (15).

Acute flares are best treated with broad-spectrum antibiotics (usually administered for 4-6 weeks), or sometimes oral zinc (15). In the majority of patients, good results are also achieved by subsequent treatment with isotretinoin. While oral antibiotics and/or systemic (or intralesional) steroids have been required for nodular scarring in the third stage of the disease, oral isotretinoin has been proven sufficiently efficient as monotherapy, in the abscess-alopecia second stage. Oral isotretinoin is effective, although therapy usually needs to be continued for 4 months after clinical control is achieved, to prevent relapses (15). According to some authors, isotretinoin therapy has to last for at least 8-10 months for perifolliculitis capitis treatment. Thereby, isotretinoin has been found to have numerous good effects on pilosebaceous units, such as changes of follicle keratinization, uproot comedones, anti-inflammatory effects, and significant reduction in the number of certain bacteria (propionibacteria and micrococci) (6).

It is necessary to take care of bacterial infections in patients with perifolliculitis capitis, as they most likely represent a secondary change in the course of the disease (14). Without treatment, the outcome of the disease is scarring alopecia and, in rare cases, squamous cell carcinoma. Our case of patient with perifolliculitis capitis emphasizes that isotretinoin remains the most effective treatment that promotes long-term control of the disease.

There is also a possibility of therapy with local isotretinoin (in the form of topical retinoic acid or adapalene), which is useful for its anti-inflammatory, desquamative and keratolytic actions; in these patients, it ensured successful control of perifolliculitis capitis and prevented evolution to scarring alopecia and nodule formation (15).

Systemic corticosteroids (prednisone 40-60 mg/day/tapered off over 3 weeks) may interrupt the inflammatory response and allow for more rapid healing. On the other hand, systemic or intralesional steroids provide only partial relief, moreover, they may favor the possible malignant transformation.

In recent years, advantages of therapy with TNF-blocking agents (adalizumab) have been reported (16).

CONCLUSION

Identification of the infective agent, repeated swabs and KOH examination/or fungal cultures and tissue sampling for histopathologic analysis are often necessary to confirm the diagnosis of perifolliculitis capitis abscedens et suffodiens. In our case, a combination of oral antibiotics and antimycotics with local skin care, followed by a long period of oral isotretinoin, led to resolution and thus inhibited the evolution to scarring and nodular stage of the disease. Thus, such combined approach could be useful for other patients with the same dermatologic problems.

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