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# Perifolliculitis Capitis Abscedens et Suffodiens Treated with Systemic Isotretinoin Monotherapy: Case Report and Review of Current Therapeutic Options

*Efstathios Rallis[1], Constantinos Verros[2], Alexandros Katoulis[3]*

[1]Department of Biomedical Sciences, University of West Attica, Athens, Greece; [2]Dermatologist-Venereologist, Tripolis, Greece; [3]2nd Department of Dermatology, Attikon General University Hospital, Athens, Greece

**Corresponding author:**
Efstathios Rallis, MD
Department of Biomedical Sciences
University of West Attica
Athens, Greece
*efrall@otenet.gr*

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**ABSTRACT** Perifolliculitis capitis abscedens et suffodiens (PCAS) is a rare, suppurative dermatosis of the scalp, the etiology of which remains unknown. It is characterized by the development of comedones, perifollicular pustules, firm or fluctuant and itchy or painful nodules and abscesses of the scalp, connected by communicating sinuses that may lead to the formation of scarring and irreversible alopecia. Treatment of PCAS is challenging, often leading to unsatisfactory results. We present a case of a 23-year-old Caucasian man with PCAS who was treated successfully with systemic isotretinoin monotherapy and we review the current therapeutic options.

**KEY WORDS:** perifolliculitis capitis abscedens et suffodiens, dissecting cellulitis of the scalp, isotretinoin, treatment, infliximab, adalimumab, review

## INTRODUCTION

Perifolliculitis capitis abscedens et suffodiens (PCAS) also called dissecting cellulitis of the scalp, acne necrotica miliaris, or proprionibacterium folliculitis, is a rare, suppurative dermatosis of the scalp, the etiology of which remains obscure. It occurs predominantly in men of African-American origin between 20 and 40 years of age but it has also been described in other races and in women or girls (1-4).

Clinically, PCAS is characterized by the development of different sizes of comedones, perifollicular pustules, firm or fluctuant and itchy or painful nodules and abscesses of the scalp, connected by communicating sinuses that may lead to the formation of scarring and irreversible alopecia. The most common affected areas of the scalp are the vertex and the occipital part (1). Treatment of PCAS is challenging, often leading to unsatisfactory results (5).

We present a case of PCAS treated successfully with systemic isotretinoin monotherapy and we review the current therapeutic options.

## CASE REPORT

A 23-year-old Caucasian man was referred to our clinic because of a 5-month history of painful cysts, fluctuating nodules, abscesses, and hair loss involving the occipital and vertex areas of the scalp (Figure 1), significantly affecting the patient's cosmetic appearance. If pressed, a purulent secretion was excreted from fluctuant nodules, not only from the pressed lesion but also from distant nodules. Cervical lymphatic nodes were not detected. The patient's past medical history was unremarkable, and his general health was good.

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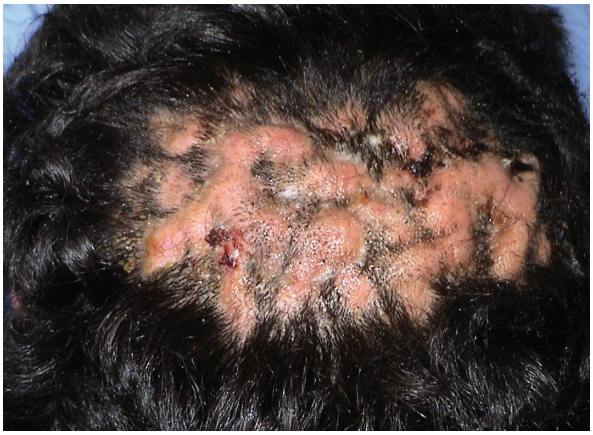

**Figure 1.** Painful cysts, fluctuating nodules, abscesses and hair loss in the occipital and vertex areas of patient's scalp.

The patient was initially unsuccessfully treated by his internist with amoxicillin with clavoulanic acid 1 gr twice daily for ten days in combination with topical application of mupirocin ointment.

The histopathological examination of an incisional biopsy sample obtained from a scalp lesion showed a dense, mixed inflammatory infiltrate consisting of neutrophils, lymphocytes, histiocytes, and plasma cells. Abscess formation was also seen in the dermis and subcutaneous tissue. Direct microbiologic examination and cultures obtained from a purulent nodule were positive for *Staphylococcus aureus*. Mycological examination was negative. Blood tests including peripheral blood picture and biochemical tests were within normal ranges. Based on these findings, a diagnosis of PCAS was established.

The patient was treated with oral isotretinoin 1 mg/kg as monotherapy for 5 months with excellent results and without any residual scarring (Figure 2). No incision of cystic lesions was performed. No relapse was observed during a 6-month follow-up (Figure 3).

## DISCUSSION

PCAS was first reported in 1903 by Spitzer (6), while Hoffman (7) named the dermatosis "perifolliculitis capitis abscedens et suffodiens" in 1908. The term "suffodiens" derives from the Latin word "suffodio", meaning to "dig under" (8).

The disease can occur alone or as a part of the follicular occlusion tetrad, which also includes acne conglobate, hidradenitis suppurativa, and pilonidal cysts (9).

The participation of PCAS in this tetrad may suggest a pathogenic mechanism involving deep follicular occlusion, accumulation of material in the follicle, dilatation of the follicle, follicular rupture, and subsequently follicular infection (2,10). It seems that a primary immunological response occurs, characterized by the early appearance of lymphocytes, commencing from the bacterial and keratin structures that are released from the ruptured follicles, with secondary bacterial infection commonly caused by *Staphylococcus aureus*, *Staphylococcus epidermidis*, or *Staphylococcus albus* (2,8). The TNF-α levels in the serum and/or the skin have not been reported in patients with PCAS; however, the involvement of TNF-α is assumed due to the frequent coexistence of PCAS and hidradenitis suppurativa. Although the exact cause of PCAS still remains unknown, the onset at a young age, the increased incidence in man and in individuals with a dark phototype, and the few cases of familiar PCAS reported in the literature (11) suggest a genetic and/or a hormonal risk factor (10).

Serious complications of this dermatosis include the development of osteomyelitis of the skull under lesions of dissecting cellulitis (12) and squamous cell carcinoma arising from scarring, chronic relapsing lesions (13). However, no SCC was recorded in retrospective study by Badaoui *et al.* on 51 patients with PCAS (10). In the same study, 12% of the patients had hidradenitis suppurativa, 16% had acne conglobata, and 4% had both acne conglobata and hidradenitis suppurativa, while none of the patients had pilonidal cyst, psoriasis, inflammatory bowel disease, or inflammatory arthritis. In this series, only a few patients had a disease associated with the follicular tetrad.

PCAS is an uncommon, chronic, and relapsing disease with an unpredictable course, showing considerable resistance to conservative management. Several treatments have been used in the literature, most of them with poor results.

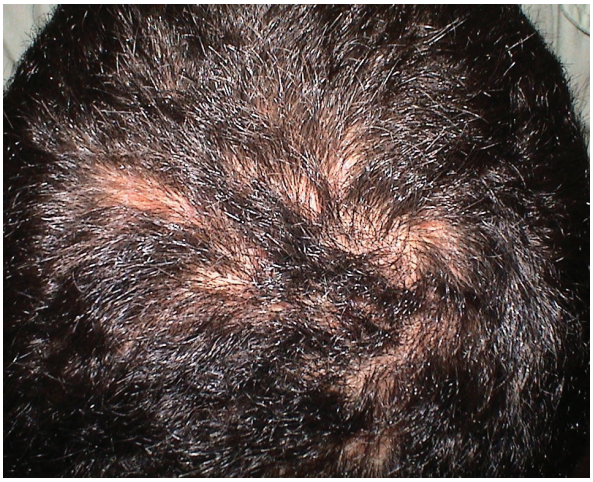

**Figure 2.** The patient was treated with oral isotretinoin as monotherapy for 5 months with excellent results.

Based on our experience and according to literature, oral isotretinoin is considered the treatment of choice for PCAS (1,8,10,11,14-19). It can be administered as monotherapy or in combination with systemic antibiotics, antimycotics, glycocorticoids, zinc, and surgical procedures (1,10,20-24). The most effective regimen is a daily dose of oral isotretinoin as monotherapy, 0.8-1.0 mg/kg/day for approximately 4-5 months (or 0.5-0.8 mg/kg/day for 5-7 months). Complete remission of the disease can be achieved in 92% of patients (10). Ljubojevic *et al.* (25) described an isotretinoin, low-dose treatment regimen for PCAS, starting with an initial dose of 30 mg and continuing with 10 mg daily over 10 months. Relapses are frequent after discontinuation of treatment, but no recurrence was observed in our case, six months after the completion of isotretinoin intake.

Other retinoids, such as oral acitretin (26) and alitretinoin (27), have been reported as PCAS treatment. Successful control (but not treatment) of the disease, with interruption of the evolution of PCAS to scarring alopecia and nodule formation, has been described in only one case (28).

Zinc supplementation and antibiotics including doxycycline, metronidazole, clindamycin, pristinamycin, rifampicin, and antibiotic soaps (chlorohexidine, benzoyl peroxide), have been used for the treatment of PCAS either as monotherapy or as a combination with oral isotretinoin, with moderate improvement observed in all patients; however, the patients relapsed after discontinuation of treatment (1,8,10,20-22,29).

Adalimumab and infliximab have been also administered for the treatment of PCAS (10, 29-37); however, data are limited. In most cases, anti-TNF agents showed significant improvement in PCAS with no concomitant effect on fibrosis or cicatrization.

Recently, photodynamic therapy (PDT) combined either with surgery (38,39) or with pre-treatment by fire needle intervention with isotretinoin (40) demonstrated safety and showing satisfactory results.

## CONCLUSION

Treatment of PCAS still remains challenging. Systemic isotretinoin is currently the most effective treatment, but relapses were observed. TNF-α inhibitors seem to represent a promising therapeutic option, but data on their efficacy and the recurrence rate of PCAS are limited.

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