

CASE REPORT

Alopecic and aseptic nodules of the scalp: A new case with a systematic review of the literature

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Abstract

Alopecic and aseptic nodule of the scalp/Pseudocyst of the scalp is a rare but probably underdiagnosed nonscarring alopecia with good prognosis and doxycycline is a safe and effective option treatment.

KEYWORDS

alopecia, hair, nodules, pseudocyst, scalp

1 | INTRODUCTION

Alopecic and aseptic nodule of the scalp (AANS) is a rare nonscarring inflammatory alopecia presenting in young males with alopecic dome-shaped nodules without any microbial colonization. We report a case of AANS in a 39-year-old man and a summary of AANS clinicopathological characteristics, course, and treatments, according to the best evidence.

Alopecic and aseptic nodule of the scalp is a rare disease characterized by a solitary nodule or few nonscarring alopecic nodules, without any microbial colonization. AANS presents with dome-shaped, firm, or soft in consistence nodules of the scalp that when punctured may drain a sterile citrine, purulent or blood-tinged fluid. Histopathology shows a nonspecific inflammatory infiltrate in the deep dermis, however a granulomatous infiltrate mixed with lymphocytes and histiocytes or pseudocyst formation may be found. The disease course may last few months, but generally follow a benign prognosis.^{1,2} To our knowledge, more than 70 cases have been described. We report the clinical and histopathologic features of a new case of AANS in a young man and a systematic literature review to provide a summary of the clinical

presentation and the effective treatments for AANS, according to the best evidence available from published studies.

2 | METHODS

2.1 | Case report

A 39-year-old man presented with an alopecic nodule of the scalp from 1 year. No subjective symptoms as itch or pain were reported. Clinical examination revealed a 15 mm, dome-shaped alopecic nodule in the occiput surrounded by normal appearing scalp tissue (Figure 1A,B). The consistence was soft and mobile, and the puncture drained a sterile citrine liquid. Histological examination showed in a background of normal follicular count a circumscribed perifollicular inflammation in the deep dermis. It was composed by neutrophils, plasma cells, and lymphocytes with focal granuloma formation (Figure 1C,D). Specific stains ruled out bacteria and fungi. No drugs were administered as the patients obtained a complete remission following the biopsy with hair regrowth in 2 months. Six months after the remission, the patient had no relapses.

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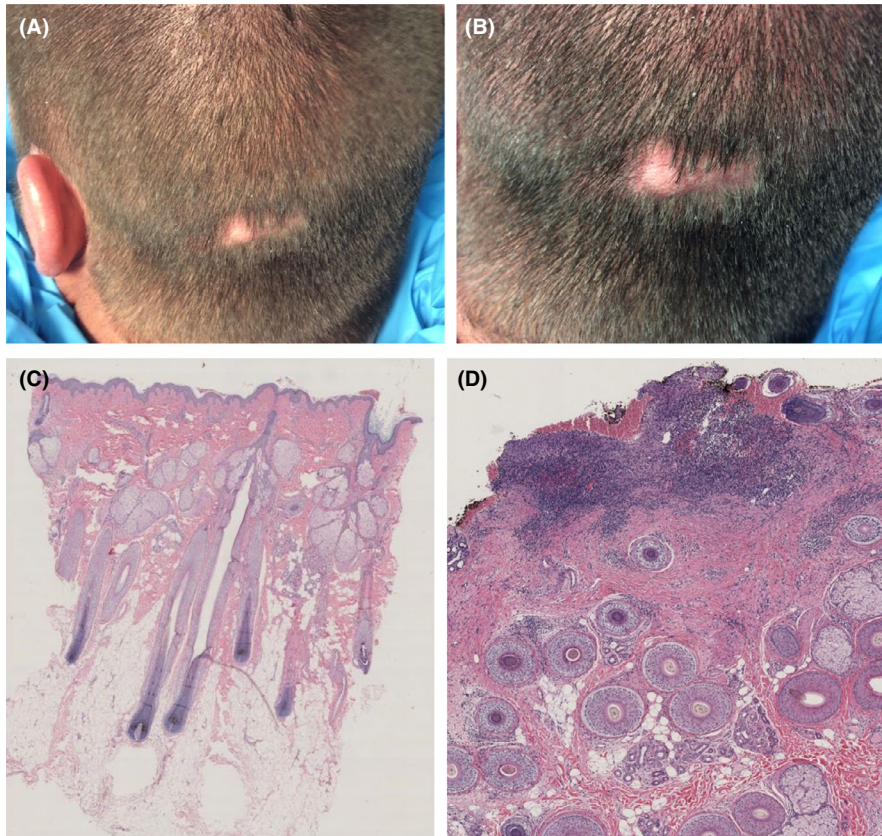


FIGURE 1 Single dome-shaped alopecic skin-colored nodule of the occiput in a 39-y-old man (A, B). Vertical (40 \times ; C) and horizontal sections (100 \times ; D) showing a circumscribed perifollicular inflammation in the deep dermis composed by neutrophils, plasma cells, and lymphocytes with focal granuloma formation (hematoxylin eosin stain)

2.2 | Literary review

Electronic searches on PubMed[®] and Ovid Medline[®] database employing “alopecic aseptic nodules scalp” OR “pseudocyst scalp” as search term were performed. Original articles published until February 1, 2021 and describing clinical and histological characteristics and treatments of AANS/pseudocyst of the scalp were included. Articles written in a language other than English were excluded. Articles were screened by title and abstract and those deemed relevant were reviewed in full text and selected or rejected based on the inclusion and exclusion criteria. The inclusion/exclusion criteria were applied also for the references of the articles deemed relevant. Data concerning items were extracted as follows: age and sex of the patients, number, size, consistence and site of scalp lesions, symptoms, disease duration, characteristic of fluid at the puncture, trichoscopic, ultrasonography and histopathological features, therapeutic interventions, clinical outcome considering complete or partial hair regrowth.

3 | RESULTS

A total of 25 articles were retrieved through electronic database searches and were screened by title and abstract. A number of 15 original reports meeting the inclusion and exclusion criteria were analyzed, including eight case reports

and seven case series. As shown in Table 1, in addition to the present case, a total of 71 patients with AANS/pseudocyst of the scalp were identified, 43 (60%) men and 29 (40%) women. The mean age was 27.1 years (range 11–72). The mean duration of disease prior examination was almost 1 year (mean 361 days), ranging from a week to some years. Most cases presented with one or two (mean 1.5) alopecic dome-shaped nodules with a mean diameter of 2.1 cm (range 1.2–5 cm). The nodules were generally firm in consistence (67%), although also fluctuating (22%) and soft (11%) nodules were reported. The commonest scalp site was the vertex (n = 26; 51%), followed by occiput (n = 18; 35%), parietal (n = 4; 8%), and temporal (n = 2; 4%) scalp. One case presented with beard involvement. Most cases were completely asymptomatic (63%) but in more than one third of the cases, itching or pain was reported (37%). Trichoscopic examination was assessed in four cases, all presenting with black and yellow dots, fine vellus, and broken hairs. In other four cases, ultrasonography was performed, describing hypoechoic or anechoic subcutaneous nodular lesions. Histopathology show generally a mixed or granulomatous inflammatory cell infiltrate in the mid and/or deep dermis composed by lymphocytes, neutrophils, plasma cells, and histiocytes sometimes along with multinucleated giant cells and granulomas. In nine cases, pseudocyst formation was found. Doxycycline 100–200 mg/d for 8–12 weeks was the most used treatment,

TABLE 1 Clinical, histopathologic features, and treatments of the cases of AANS reported in the literature

Cases	N = 71
	Case report (n = 8) Case series (n = 7)
Age (y), mean ± SD	27.1 ± 9.5 (range 11-72).
Male, n (%)	43 (60)
Disease duration (d)	361 ± 352
Number of nodules, mean ± SD	1.5 ± 1
Nodule diameter (cm), mean ± SD	2.1 ± 0.8
Nodule consistence, n (%)	Firm; 24 (67) Fluctuating; 8 (22) Soft; 4 (11)
Location, n (%)	Vertex; 26 (50) Occiput; 18 (35) Parietal; 4 (8) Temporal; 2 (4) Beard area; 1 (2)
Symptoms, n (%)	Asymptomatic; 33 (64) Itch or pain; 19 (37)
Trichoscopic features	Black and yellow dots, fine vellus, and broken hairs (four cases reported)
Ultrasonography features	Hypoechoic or anechoic subcutaneous nodular lesions (five cases reported)
Histopathology	Mixed or granulomatous inflammatory cell infiltrate in the mid and/or deep dermis composed by lymphocytes, neutrophils, plasma cells, and histiocytes sometimes along with multinucleated giant cells and granulomas. In nine cases, pseudocyst formation was found.
Treatment	Doxycycline 100-200 mg/d for 8-12 wk; 24 cases Intralesional corticosteroids (multiple triamcinolone acetonide injections); 14 cases Oral cephalosporine; 10 cases
Outcome, n (%)	Complete remission with hair regrowth; 32 (89) Partial hair regrowth was reported; 4 (11)

Abbreviations: AANS alopecic and aseptic nodules of the scalp; SD, standard deviation.

followed by intralesional corticosteroids (multiple triamcinolone acetonide injections) and oral cephalosporine. Although the outcome was reported only in half analyzed

cases, most cases (89%) had complete remission with hair regrowth. In four cases, partial hair regrowth was reported.

4 | DISCUSSION

Alopecic and aseptic nodule of the scalp is a rare inflammatory nonscarring alopecia affecting mainly young males and presenting with one or few alopecic dome-shaped nodules surrounded by normal scalp. The lesions may be firm, soft, or fluctuating in consistence and generally are asymptomatic or associated with itch or pain. The nodules were commonly found in the vertex or the upper part of the occiput. The staining and the microbiological cultures for fungi and bacteria were always negative.^{1,2} There is an increasing number of reports describing AANS suggesting that this entity may be not so rare and probably underdiagnosed.³⁻⁹ A similar nosological entity, known as pseudocyst of the scalp (PCS), in different Asian case reports was previously described.^{10,11} It is unclear whether PCS and AANS represent the same spectrum given some differences in the clinical presentation and histopathology.² The name AANS was coined by Abdennader and Reygagne in 2009^{11,12} in a case series of 18 cases, all lacking the pseudocyst-like architecture. The cases reported in the Japanese literature described as PCS generally presented with alopecic nodules between the top and the forehead of the scalp, when punctured the drainage liquid was citrine-colored or blood-tinged and histopathology showed a pseudocyst-like architecture with a granulomatous infiltration. By contrast, AANS generally presented with alopecic nodules in the vertex and upper part of the occiput, when punctured the drainage liquid might be purulent and histopathology showed a nonspecific mixed infiltration and in most cases granuloma in the deep dermis. These differences may be due to racial variances in the type of hair, the timing and site of biopsy, while the type of puncture discharge may depend on the predominant type of the infiltrate and the presence of eroded vessels.

The differential diagnosis included mainly dissecting cellulitis of the scalp (perifolliculitis capitis abscedens et suffodiens), a scarring alopecia presenting with tract sinus, trichilemmal cyst that is not associated with alopecia, scalp cutaneous metastasis and primary cutaneous B-cell lymphoma.² Cutaneous metastasis present with alopecic, skin-colored and firm plaques or nodules generally derived from breast cancer in women and lung or renal cancer in men. Primary cutaneous B cells lymphoma show alopecic, erythematous or pink-violet, firm plaques and nodules that rarely ulcerate.

Trichoscopy and ultrasonography may help for the diagnosis.¹³⁻¹⁵ Trichoscopy features include broken hair, black dots, yellow dots, and fine vellus. The “Eastern pancake sign” characterized by dilated follicular openings has been

described. Ultrasonography showed well-defined subcutaneous hypoechoic nodules lacking the heterogeneous center with hyperechoic lines that characterize the trichilemmal cyst.

The most employed treatment is a 3 month course of doxycycline 100-200 mg/d, but spontaneous resolution following drainage puncture or biopsy was frequently described. Other options included intralesional corticosteroids injections, surgical excision, and oral cephalosporin. Although the disease course is long, the prognosis is good and a complete remission with hair regrowth occurred in almost all the patients.

The etiology is still debated: a deep folliculitis without bulge involvement and causing a nonscarring alopecia has been proposed. Another hypothesis considered AANS in the spectrum of follicular occlusion diseases such as hydro-sadenitis suppurativa and acne conglobata.² Further studies are needed to better clarify the etiopathogenesis and the best treatment for AANS.

In conclusion, AANS/PCS is a rare but probably underdiagnosed nonscarring alopecia characterized by alopecic nodules of the scalp in absence of any bacterial or fungal infection. Given the good prognosis a brief course of doxycycline is generally a safe and effective option treatment.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

FB: contributed to data curation, investigation, methodology, software, and writing—original draft. MM: involved in conceptualization, data curation, and formal analysis. CC, PG, and GG: involved in supervision, validation, visualization, and writing—review and editing. All the authors approved the submitted version.

CONSENT STATEMENT

Published with written consent of the patient.

DATA AVAILABILITY STATEMENT


The data that support the findings of this study are available from the corresponding author upon reasonable request.

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How to cite this article: Bellinato F, Maurelli M, Colato C, Gisondi P, Girolomoni G. Alopecic and aseptic nodules of the scalp: A new case with a systematic review of the literature. *Clin Case Rep*. 2021;9:e04153. <https://doi.org/10.1002/ccr3.4153>