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Alopecia areata after DRESS syndrome with a rapid resolution

Alopecia areata induzida após síndrome DRESS com rápida resolução

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ABSTRACT

Anagen effluvium (AE) is a hair growth disorder that occurs due to a disturbance in the hair follicle cycling. The most common cause of AE is alopecia areata (AA). Autoimmune diseases like AA may develop after drug reaction with eosinophilia and systemic symptoms (DRESS) syndrome and are usually long-standing conditions. We report the case of a 17-year-old woman who developed a severe and abrupt AE after DRESS syndrome with a full and rapid recovery after two months of topical minoxidil.

Keywords: Alopecia; Alopecia Areata; Drug Hypersensitivity Syndrome; Case Reports

RESUMO

O eflúvio anágeno (AE) é um distúrbio no ciclo do folículo piloso, cuja causa mais comum é a alopecia areata (AA). Doenças autoimunes, como a AA, podem manifestar-se após a síndrome de reação a drogas com eosinofilia e sintomas sistêmicos (DRESS) e, em geral, cursam com sinais e sintomas de longa duração. Relatamos o caso de uma paciente de 17 anos que desenvolveu um quadro abrupto de AE, após o diagnóstico de síndrome DRESS, e que apresentou rápida e completa recuperação capilar após uso de minoxidil tópico por dois meses.

Palavras-chave: *Alopecia; Alopecia em áreas; Síndrome de hipersensibilidade a medicamentos; Relatos de casos*

Case Report

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INTRODUCTION

Anagen effluvium (AE) is a diffuse, non-scarring alopecia characterized by the loss of anagen hairs as an acute insult result. The most common cause of AE is alopecia areata (AA). Drug reaction with eosinophilia and systemic symptoms (DRESS) presents with severe skin rash, fever, lymphadenopathy, and multiple organ involvement. Autoimmune diseases, such as AA, can manifest after DRESS syndrome and, in general, present long-lasting signs and symptoms.[1] The mechanisms of this condition are not fully understood, and the development of autoimmune disorders after DRESS syndrome involves the activation of T lymphocytes, with possible viral reactivation.[2]

CASE REPORT

A 17-year-old woman was admitted to the hospital due to DRESS syndrome caused by the use of amoxicillin with potassium clavulanate. She received systemic corticosteroids, plasmapheresis, and cyclophosphamide. The patient was discharged after the resolution of the condition, using only prednisone 5 mg daily, suspended after a week. Ten days after hospital discharge, the patient presented extensive AE (Figure 1). Trichoscopy revealed diffuse capillary rarefaction, black dots, yellow dots, and dystrophic anagens (Figure 2). The scalp biopsy demonstrated a peribulbar lymphocytic infiltrate, supporting the diagnosis of AA (Figure 3). After two months of using topical minoxidil without systemic treatment, the patient showed complete hair recovery and no longer had signs of disease activity (Figure 4).

Figure 1: Diffuse hair loss with the presence of thin, short, sparse strands

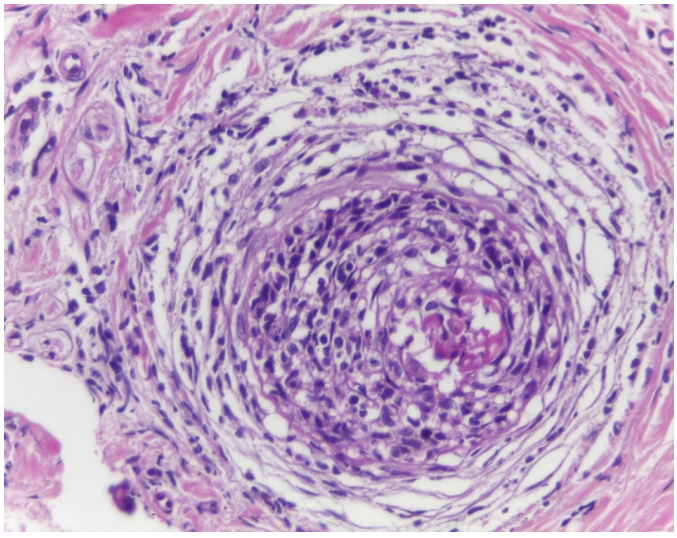

Figure 3: Peribulbar lymphocytic infiltrate similar to a "swarm of bees" around the follicle in the anagen phase

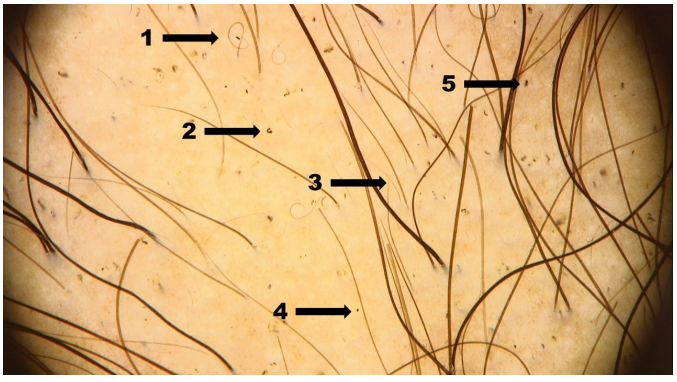

Figure 2: Trichoscopy demonstrates reduction in hair density, hair shafts of varying thickness, circular hairs (1), yellow dots with remaining amorphous hairs (2), fine and short hairs (3), black dots (4) and fractured hairs (5)

Figure 4: Complete hair recovery after topical use of 5% minoxidil for two months

DISCUSSION

AE is a consequence of an autoimmune inflammatory process against the hair follicle bulb in the anagen phase, which interrupts the follicle's mitotic activity. This phenomenon is primarily observed in cases of AA.[1]

Autoimmune diseases, such as graft versus host disease, hemolytic anemia, vitiligo, alopecia areata, systemic lupus erythematosus, thyroid diseases, type 1 *diabetes mellitus*, rheumatoid arthritis, and hypersensitivity to new drugs, may develop after the resolution of DRESS syndrome.[3] Cases of AA that occur after DRESS syndrome are, in general, long-lasting (Table 1).[4,5,2,6,7,7,8]

Table 1: Cases of alopecia after DRESS syndrome

Reference	Cases	Age/Sex	Diagnosis	Responsible drug	Onset of symptoms after drug introduction	Hair growth	Clinical findings	Trichoscopic findings	Histopathological findings
Retrospective cohort									
Chen 2013	1	27/M	Alopecia areata	Ampicillin	2 years (after DRESS syndrome)	Unavailable	Patchy, recurring hair loss on the scalp, eyebrows and thighs	Unavailable	Unavailable
Retrospective									
Ushigome 2013	1	45/W	Alopecia	Unavailable	4 months	Unavailable	Unavailable	Unavailable	Unavailable
Case report									
Hollingsworth 2020	1	14/W	Alopecia universal	Lamotrigine	2 months	No evidence of hair growth after 9 months of methotrexate use	Complete hair loss on the scalp with preservation of the eyebrows and eyelashes	Unavailable	Unavailable
Morita 2018	1	6/M	Alopecia Alopecia areata and alopecia totalis	Sulfamethoxazole - trimethoprim Zonisamide	7 months: alopecia areata 21 months: alopecia totalis 5 months	Unavailable	Unavailable	Unavailable	Unavailable
Morita 2018	1	17/W	Alopecia				Unavailable	Unavailable	Unavailable
Lan 2016	1	13/W	Alopecia	Minocycline	10 months	Unavailable	Unavailable	Unavailable	Unavailable
Kano 2007	1	46/W	Diffuse alopecia	Zonisamide	11 months	No improvement in alopecia was observed after 5 months of follow-up	Diffuse scalp alopecia	Unavailable	Unavailable
Our case	1	17/W	Alopecia areata	Amoxicillin + Clavulanate	1 month	Complete recovery after 2 months of topical use of minoxidil 5%	Diffuse and acute anagen effluvium on the scalp	Black dots, yellow dots and dystrophic anagens	Non-scarring alopecia with an increase in the number of follicles in catagen and telogen phases. Miniaturized follicles, anagen hairs and peribulbar lymphocytic infiltrate

W, woman; M, man; DRESS – drug reaction with eosinophilia and systemic symptoms.

The reaction to drugs alone could represent a breakdown in the immune system's tolerance and be a risk factor for autoimmunity development. DRESS syndrome can be considered as part of an autoimmune spectrum.[3]

AA and DRESS syndrome are conditions mediated by T lymphocytes, possibly of autoimmune origin, whose mechanisms are not fully understood. The development of autoimmune diseases after the cure of DRESS syndrome may be related to the change in the expression of regulatory T lymphocytes (Tregs) and a gradual loss of function of these lymphocytes.[9]

Well-known factors in the development of AA, such as immune privilege collapse, genetic inheritance, the role of the ULBP genes and the ULBP3 and NKG2D receptors, and innate and acquired immunity involvement, also appear to participate in the pathophysiogenesis of DRESS syndrome.[9,10]

The episode of hair loss described in our patient occurred just one month after the development of DRESS syndrome and was quickly resolved with topical minoxidil as monotherapy. To our knowledge, this is the first case report describing rapid recovery from AA triggered after DRESS syndrome resolution.

CONCLUSION

The reported case illustrates the development of AA, an autoimmune disease, after DRESS syndrome resolution. We describe for the first time a patient who had complete and rapid AA resolution using only topical treatment. The immunological mechanism involved remains to be elucidated.

REFERENCES:

1. Kanwar AJ, Narang T. Anagen effluvium. Indian J Dermatol Venereol Leprol. 2013;79(5):604–12.
2. Hollingsworth P, Paci K, Evans M, Miedema J, Morrell DS. Alopecia universalis after drug reaction with eosinophilia and systemic symptoms (Dress). Pediatr Dermatol. 2020;37(5):947–9.
3. Sasidharanpillai S, Joseph AT, Ajithkumar K, Devi K. Autoimmune diseases, end organ dysfunction and adverse drug reaction following drug reaction with eosinophilia and systemic symptoms (DRESS): a retrospective cohort study. Indian Dermatol Online J. 2021;12(5):722-5.
4. Chen YC, Chang CY, Cho YT, Chiu HC, Chu CY. Long-term sequelae of drug reaction with eosinophilia and systemic symptoms: a retrospective cohort study from Taiwan. J Am Acad Dermatol. 2013;68(3):459–65.
5. Ushigome Y, Kano Y, Ishida T, Hirahara K, Shiohara T. Short- and long--term outcomes of 34 patients with drug-induced hypersensitivity syndrome in a single institution. J Am Acad Dermatol. 2013;68(5):721–8.
6. Morita C, Yanase T, Shiohara T, Aoyama Y. Aggressive treatment in paediatric or young patients with drug-induced hypersensitivity syndrome (DiHS)/drug reaction with eosinophilia and systemic symptoms (DRESS) is associated with future development of type III polyglandular autoimmune syndrome. BMJ Case Rep. 2018;2018:bcr2018225528.
7. Lan J, Lahoti A, Lew DB. A severe case of minocycline-induced DRESS resulting in liver transplantation and autoimmune sequelae. Ann Allergy Asthma Immunol. 2016;116(4):367–8.
8. Kano Y, Sakuma K, Shiohara T. Sclerodermoid graft-versus-host disease--like lesions occurring after drug-induced hypersensitivity syndrome. Br J Dermatol. 2007;156(5):1061–3.
9. Shiohara T, Mizukawa Y. Drug-induced hypersensitivity syndrome (DiHS)/drug reaction with eosinophilia and systemic symptoms (DRESS): an update in 2019. Allergol Int. 2019;68(3):301–8.
10. Speiser JJ, Mondo D, Mehta V, Marcial SA, Kini A, Hutchens KA. Regulatory T-cells in alopecia areata. J Cutan Pathol. 2019;46(9):653–8.

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