

Case Report

A rare case of azathioprine induced anagen effluvium and plica neuropathica

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Received: 26 February 2024

Revised: 22 March 2024

Accepted: 27 March 2024

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ABSTRACT

Anagen effluvium is a type of hair loss that results from a sudden decrease in the metabolic activity of the hair follicle's matrix cells. The condition is commonly associated with cancer treatments such as chemotherapy and radiation, but can also be caused by other factors. Plica neuropathica, on the other hand, is a condition where hair on the scalp becomes irreversibly matted. It is often linked to psychiatric disturbances, poor hair care, scalp infestations, and the use of ionic surfactants in shampoos. In rare cases, it has also been associated with certain medications. Here we present a rare case of a 16-year-old female who was on azathioprine and was diagnosed with an overlap of both conditions.

Keywords: Anagen effluvium, Azathioprine, Plica neuropathica

INTRODUCTION

Anagen effluvium refers to abrupt shedding of hair in the anagen phase due to impaired follicular mitotic activity. Anagen effluvium is often associated with chemotherapy, but it can also be caused by other factors that disrupt the normal cycle of hair growth. Common culprit drugs are chemotherapeutic agents, toxic metals and rarely bismuth, levodopa, colchicine and cyclosporine.¹ Plica neuropathica, an uncommon condition, is characterized by the irreversible tangling, twisting, and matting of hair in individuals who are otherwise healthy.^{2,3} This condition, once thought to be unique to the Poles (hence the name plica polonica), typically arises due to neglect or parasitic infestations.³ It is also known as bird's nest hair, plica polonica, or felted hair.

CASE REPORT

A 16-year-old female presented to our dermatology OPD with an abrupt onset of loss of hair over scalp in large clumps and matting of hair over occipital region since past 20 days. About a month back patient was diagnosed with chronic urticaria by a dermatologist in a different hospital and was prescribed tablet Azathioprine 50 mg twice a day for the same. Within 15 days of treatment, she developed oral ulcers which got resolved on discontinuation of azathioprine. She also noticed hair loss in the form of clumps from the frontal region of the scalp about 20 days back. Due to which the patient had stopped the use of a hair comb and gradually also stopped shampooing her hair. There was no history of tenderness or pain over the scalp, itching, or malodour from the scalp. She was otherwise healthy and had no history of

any concomitant psychiatric disorder. No history of pediculosis capitis or any ectoparasitic infection could be elicited. There was no history of use of any chemical treatment for her hair like streaking or straightening of hair in the past. On examination of the scalp, diffuse non-scarring alopecia was seen more on the frontal region than on the temporal and the parietal regions (Figure 1-2).



Figure 1: Diffuse non scarring alopecia of the frontal and bilateral parietal and bilateral temporal region.



Figure 2: Diffuse non scarring alopecia of the occipital region.

There was also a mass of matted entangled hair over the vertex area extending up to the occipital region (Figure 3). There were no signs of inflammation, scaling, atrophy or nits. On performing a hair pull test, hair came off in massive clumps with relatively little effort over the frontal region of the scalp. On trichoscopy, multiple black dots with few short regrowing hairs, circle hairs, yellow dots and flame hairs were observed (Figure 4), suggestive of anagen effluvium. All routine investigations were within normal limits. Naranjo ADR Probability Scale was used and causality assessment score was 6 indicating that azathioprine was a “probable” cause of the anagen effluvium. Furthermore, the patient was advised to discontinue the use of Azathioprine immediately. The patient was then prescribed oral hair growth supplements,

topical Procapil and Redensyl solution, and topical Minoxidil 5% lotion once daily. Patient reported some improvement and hair growth on follow-up after 1 month but then was lost to follow up.



Figure 3: A mass of dense entangled hair giving bird's nest appearance over vertex region.

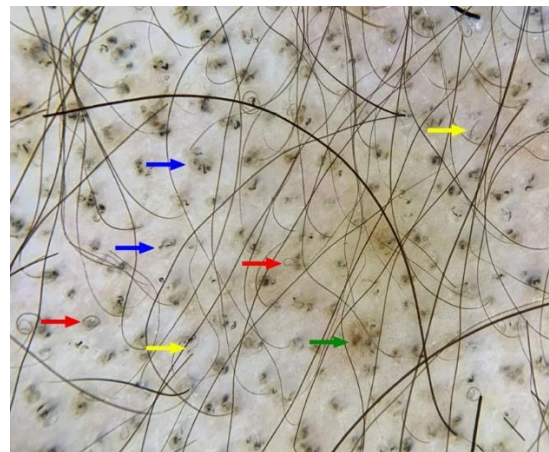


Figure 4: Trichoscopy: showing multiple black dots and few circle hair (red arrow), yellow dots (blue arrow), dark line (yellow arrow), and flame hair (green arrow) (Dermlite DL4, 10X magnification, contact mode).

DISCUSSION

Human hair typically goes through three stages in its growth cycle: anagen, catagen, and telogen. The anagen phase is when the hair is actively growing, and this is when the matrix cells of the hair follicle are highly active in mitosis. On an average scalp with 100,000 hairs, about 10-15% of them are in the catagen or telogen phase at any given moment. However, the majority of hair follicles are in the anagen stage.⁴ Anagen effluvium is a condition where there is a sudden loss of hair that is in its growing phase (anagen). Chemotherapeutic agents, which are commonly used for systemic cancer treatment, are the primary culprits. These agents, including antimetabolites

and alkylating agents among others, function in various ways. However, they all share a common mechanism - they inhibit the metabolism of rapidly dividing cells in the hair matrix.¹ Other medications that can rarely cause anagen effluvium include bismuth, levodopa, colchicine, and cyclosporine.¹ Hair loss typically starts 1 to 3 weeks following a certain event. The scalp, due to its prolonged anagen phase, is the most frequent site of hair shedding. The impact on other terminal hairs varies, depending on the proportion of hairs in the anagen phase. It is usually reversible within 1 to 3 months after stopping the offending agent.⁵ Although rare, few cases of azathioprine-induced anagen effluvium has been reported in past.^{6,7} In 1884, Le Page coined the term “plica neuropathica” while describing a young female who suddenly developed a tangled scalp, and she had a history of hysteria.² Other terms for the same disorder include felting and bird’s-nest hair.² The exact mechanisms that cause Plica neuropathica are not known. However, it has been suggested that the condition may be due to the splitting or weathering of the hair shaft caused by vigorous friction and the frequent use of harsh shampoos and cleansers. It may also be due to poor hair care practices in individuals with long hair.⁸ Psychiatric morbidities may also result in development of Plica neuropathica due to neglect of hair, secondary infestations and religious or superstitious beliefs and a case of Plica neuropathica has also been described in a psychiatrically ill patient.³ Although there have been a few rare cases of drug-induced plica neuropathica, such as two instances in individuals with pancytopenia who were taking azathioprine, the condition was attributed to damage to the hair shaft cuticle. This was despite the fact that these individuals did not engage in poor hair care practices.^{9,10} Our patient experienced hair matting, which may have been caused by anagen effluvium. This can occur when immunosuppressive drugs, such as azathioprine, affect the cells in the hair matrix and impair the metabolic activity of hair follicles which can result in a narrowed or defective hair shaft, making the hair more susceptible to matting. Avoidance of combing and shampooing may have exacerbated the condition.

CONCLUSION

We’re documenting a case who suffered from a rare but significant side effect from Azathioprine. Azathioprine is an immunosuppressive drug used in a variety of

dermatological and non-dermatological conditions. This documentation will assist physicians and dermatologists to be more conscious and vigilant in recognizing and managing such side effects of Azathioprine.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Sperling LC. Hair and systemic disease. *Dermatol Clin.* 2001;19(4):711-26.
2. Le Page JF. On Neuropathic Plica. *Br Med J.* 1884; 1(1204):160.
3. Kumar PN, Antony B, Chakravarthy A, Koyamu AM. Plica neuropathica (polonica) in schizophrenia - a case report and review of literature. *Indian J Psychiatr.* 2001;43(3):281-3.
4. Vogt A, McElvee K, Blume-Peytavi U. Biology of the hair follicle. In: Blume-Peytavi U, Tosti A, Whiting D, Trueb R, editors. *Textbook on hair - From basic science to clinical application.* Berlin: Springer Verlag; 2008;3:1-22.
5. Kanwar AJ, Narang T. Anagen effluvium. *Indian J Dermatol Venereol Leprol.* 2013;79(5):604-12.
6. Sharma R, Poddar S, Podder I. Azathioprine Induced Anagen Effluvium- An Uncommon Adverse Effect of a Commonly Used Drug. *Indian J Dermatol.* 2022; 67(6):815-7.
7. Bhokare A. Azathioprine-induced alopecia as an early clinical marker of its myelotoxicity. *Indian J Drug Dermatol.* 2017;3(1):40-7.
8. Marshall J, Parker C. Felted hair untangled. *J Am Acad Dermatol.* 1989;20(4):688-90.
9. Kwinter J, Weinstein M. Plica neuropathica: novel presentation of a rare disease. *Clin Exp Dermatol.* 2006;31(6):790-2.
10. Joshi R, Singh S. Plica Neuropathica (Plica polonica) Following Azathioprine-induced Pancytopenia. *Int J Trichol.* 2010;2(2):110-2.

Cite this article as: Rijhwani M, Patel MD, Malik A, Nijhawan M. A rare case of azathioprine induced anagen effluvium and plica neuropathica. *Int J Res Dermatol* 2024;10:145-7.