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Childhood Alopecia Totalis: A Case and A Concise Review of The Available Evidence-Based Therapies

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1. Abstract

Background: In the modern medical literature, alopecia totalis has been reported as early as the 1940s. Childhood alopecia totalis is a heterogeneous disorder that occur in syndromic and non-syndromic forms and can have autoimmune, nutritional and genetic bases. Non-syndromic childhood alopecia totalis is generally has autoimmune bases. During the 1970s, authors reported the treatment of alopecia totalis with DNCB which is 1-chloro, 2, 4-dinitrobenzene and High-dose glucocorticoid. Early during the 1980s, oral photochemotherapy was used in the treatment of alopecia totalis without the occurrence of side effects. The aim of this paper is to present a case of childhood non-syndromic alopecia totalis and to review of the available evidence-based therapies.

Patients and methods: The initial evidence-based treatment of a ten-year old boy with non-syndromic autoimmune alopecia totalis of more than two years that was considered refractory to treatments by at least three dermatologists is described. The family was referred to the pediatric psychiatry clinic as they no longer able to take the advice of any dermatologist and the child was clearly experiencing a psychologic upset that was disturbing the family life. Although many therapies have been tried during the previous two decades, no therapy is considered effective or satisfactory. However, in this case, the decision was made to use topical tofacitinib and topical minoxidil based on the available research evidence. However, topical tofacitinib was not available immediately, and the decision was made to use paste of raw garlic topically plus topical minoxidil.

Results: The boy was seen on the 27th of June, 2019, He had unexpectedly very scanty regrowth that contributed significantly to the improvement of the psychology of the boy and his parents.

Conclusion: It can be useful to use the available research evidence to make some improvements in challenging cases.

2. Keywords: Pediatric Alopecia Totalis, Evidence-Based Treatment

3. Introduction

In the modern medical literature, alopecia totalis has been reported as early as the 1940s [1,2]. Childhood alopecia totalis is a heterogeneous disorder that occurs in syndromic and non-syndromic forms and can have autoimmune, nutritional and genetic bases. Non-syndromic childhood alopecia totalis is generally has autoimmune bases. Hereditary alopecia totalis

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occurring in a mother and daughter was reported by Schulze [3].

Senter reported a patient with an autosomal recessive syndromic alopecia totalis associated with atypical ichthyosiform erythroderma, congenital neurosensory deafness, and vascularization of the corneas progressing to blindness, abnormalities of the teeth and nails, and postnatal growth deficiency. Senter reviewed twelve other patients with the syndrome in the literature [4, 5].

Genetic syndromic alopecia totalis was also reported in association with hereditary vitamin D-resistant rickets (Abalı et al [6].

Manz et al [7] reported the association of alopecia totalis with autoimmune polyglandular syndrome (APS 1) which is a rare autosomal recessive condition in a 21-year-old female. She developed chronic mucocutaneous candidiasis at the age of two years, and alopecia totalis at the age of three years. Thereafter, she developed chronic hypoparathyroidism and autoimmune adrenal insufficiency, malabsorption syndrome and pernicious anemia occurred [7].

Hart, Hoffman, and Winbaum [5] reported the occurrence of pediatric syndromic autoimmune alopecia totalis in a 14-year-old boy shortly after a pertussis-like illness. Thereafter, he developed chronic lymphocytic thyroiditis and diffuse polyneuritis [5].

Syndromic autoimmune alopecia totalis can also occur in association with vitiligo and autoimmune thyroid disease [8]. Pavic M et al reported the occurrence of alopecia totalis in one patient as an autoimmune manifestation associated with common variable immunodeficiency [9].

Sweetman et al (1981) reported non-genetic syndromic alopecia totalis resulting from nutritional deficiency of biotin in an 11-year-old retarded boy occurring in association with intake raw eggs which contain avidin, the biotin-binding protein. The boy also had an erythematous, exfoliative dermatosis, and increased excretion of 3 methylcrotonylglycine, 3-hydroxyisovaleric acid, 3-hydroxypropionic acid, methylcitric acid, and lactic acid [10].

Mock et al [11] reported the occurrence of non-genetic syndromic alopecia totalis in three patients with biotin deficiency associated with total parenteral nutrition. The patients had also hypotonia, and developmental delay. Two patients also had the characteristic scaly periorificial dermatitis, whereas one had only an intermittent scaly rash on the cheeks and occipital scalp. The rash, alopecia, and neurological abnormalities responded dramatically to biotin therapy (100 micrograms daily in all patients) One patient received an initial larger dose of 1 mg daily for one week plus 10 mg daily for 7 weeks [11].

During the 1970s, authors reported the treatment of alopecia totalis with DNCB which is 1-chloro, 2, 4-dinitrobenzene and high-dose glucocorticoid [12 - 15]. Happle and Echternacht [12] reported the treatment of one side of the head of 26 patients with alopecia totalis, with weekly applications of DNCB, dissolved in acetone to induce mild contact dermatitis. Difference between two sides in hair growth was noted in 17 out of the 26 patients. The difference was seen mostly within 3 months [12].

Frentz and Eriksen [13] reported the treatment of ten patients with long-standing alopecia totalis with sensitization with 1-chloro, 2, 4-dinitrobenzene (DNCB). Thereafter, they were painted once weekly on a 40 X 20 mm area of the vertex with DNCB in acetone, in concentrations adjusted to the allergic response. After seven weeks, growth of hair was seen in the painted area in 3 patients and after 8 weeks all over the scalp in 3 other patients [13].

Gutschmidt reported the treatment of 14 patients with longstanding alopecia totalis with dinitrochlorobenzene (DNCB) over a period of 2 to 11 months. Hair growth was observed in nine patients without the occurrence of any side effect [14]. Lindemayr reported disappointing results associated with the treatment of patients with long-standing

autoimmune alopecia totalis with local dinitrochlorobenzene (DNCB), and no hair growth was induced in any patients [16].

Early during the 1980s, Claudy and Gagnaire reported the use of oral photochemotherapy in the treatment of alopecia totalis without the occurrence of side effects [17]. The aim of this paper is to present a case of childhood non-syndromic alopecia totalis and to review of the available evidence-based therapies.

4. Patients and Methods

A ten-year old boy (Figure-1) with non-syndromic autoimmune alopecia totalis of more than two years that was considered refractory to treatments by at least three dermatologists. Unfortunately, the family in their disappointment threw in the garbage all previous prescriptions and reports by the previous dermatologists. The family was referred to the pediatric psychiatry clinic as they no longer able to take the advice of any dermatologist and child was clearly experiencing a psychologic upset that was disturbing the family life.

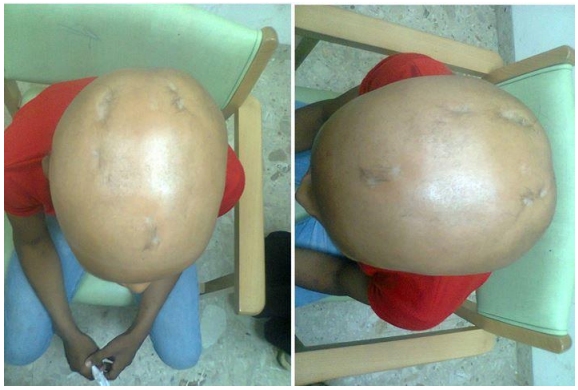

Figure 1: The boy with non-syndromic alopecia totalis of more than two years that was considered refractory to treatments by at least three dermatologists. Before receiving treatment on the 20th of June, 2020.

The family expressed that they are feeling psychologically distressed after visiting a dermatologist called Al-Sharji who claimed he was the best, but didn't prove that to them. The frustrated family mocked the name of the dermatologist saying that Al-Sharji in standard Arabic means "The anally.

5. The Therapeutic Decision

Obviously, the best therapy for the psychologic problem of the child and his family was some regrowth of hair that gives them hope. The family was reassured that we understand their frustration, and the bad news they are telling that the probably tried everything by the dermatologist. The family was told the good news which is that many patients can make improvement that is not attributed to any therapeutic agent. The family and the child didn't show much enthusiasm to this idea and waited for something better. They were assured that at the worst possibility of permanent alopecia totalis, a simple useful cosmetic solution is something like a head tattoo. The child's mood rapidly improved and was enthusiastic to that he can rapidly look as having very shortly-cut hair. Although many therapies have been tried during the previous two decades, no therapy is considered effective or satisfactory [18 - 20]. Kassira et al [18] reviewed 40 papers published during the period from the first of January, 2000, to the first of September, 2016 that tried many therapies including topical immunotherapy, steroids, photodynamic therapy, immunosuppressive agents, TNFα inhibitors, and other therapies, such as sulfasalazine, bexarotene, JAK inhibitors, and simvastatin/ezetimibe. Some treatments was associated with significant hair regrowth, no treatment was completely effective [18]. In this case, the decision was made to use topical tofacitinib and topical minoxidil based on the evidence provided by Liu, et. al, Putterman and Castelo-Soccio, Brown and Skopit, Craiglow , Shin J-W, et al, and Wambier, et al [21-26].

However, topical tofacitinib was not available immediately, and the decision was made to use paste of raw garlic topically (Based on the evidence provided by Hajheydari et al, and Hordinsky and Donati, plus topical minoxidil. The child left the paste of pure raw garlic made by his mother in the morning for at least 30 minutes, Minoxidil 5% spray in the afternoon and Minoxidil 5% gel at night.

6. Results

The boy was seen on the 27th of June, 2019, He had unexpectedly very scanty regrowth (Figure-2) that contributed significantly to the improvement of the psychology of the boy and his parents.

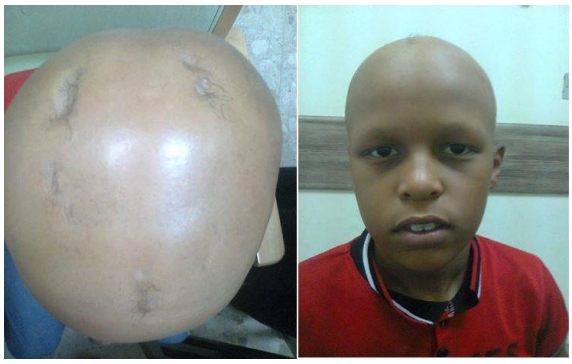

Figure 2: After treatment, had unexpectedly very scanty regrowth.

7. Discussion

Liu, et al, based on their experience with adult patients, they recommended the use of tofacitinib for the treatment of alopecia totalis, and confirmed that tofacitinib was well tolerated, and its use was not associated with serious adverse events [21].

Brown and Skopit treated satisfactorily an 8-year-old boy oral tofacitinib 5 mg twice daily with continued usage of topical steroids [23].

Shin et al described the treatment of 74 patients with refractory alopecia totalis/universalis. They treated eighteen patients with tofacitinib, and 26 patients treated with oral treatment (steroid ± cyclosporine), and 30 patients were treated with diphenylcyclopropenone. After 6 months, 44.4% of patients in the tofacitinib group, 37.5% in the oral (steroid ± cyclosporine) treatment group, and 11.1% in the diphenylcyclopropenone group achieved 50% improvements in the Severity of Alopecia Tool score. During treatment, 10% of patients in the tofacitinib group, 73.1% in the (steroid ± cyclosporine) treatment group, and 10% in the diphenylcyclopropenone group experienced adverse drug reactions. Shin et al found that oral tofacitinib was more effective than diphenylcyclopropenone immunotherapy and more tolerable than oral (steroid ± cyclosporine) treatment after six months of treatment [23 - 28].

8. Conclusion

It can be useful to use the available research evidence to make some improvements in challenging cases.

9. Acknowledgement

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10. Conflicts of Interest: None.

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