CASE REPORT

Spontaneous Re-pigmentation of Vitiligo Following Excision of Halo Congenital Melanocytic nevi: An Interesting Case Report

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Summary

Halo nevi (HN) are benign skin lesion that represent melanocytic nevi in which an inflammatory infiltrate develops, resulting in zone of depigmentation around nevus. Although Sutton originally described the lesion in 1916 as leukoderma acquista centrifugum, the lesions were noted earlier as evidenced in the painting by Matthias Grunwald cica 1512-1516. The prevalence of HNs in the general population is 1%, and HNs usually appear in childhood or early adulthood. Up to 26% of patients with HN have vitiligo, but in very few instances is there an association of HN around congenital melanocytic nevi (CMN) and vitiligo. The exact mechanisms responsible for the development of vitiligo and HN and its resolution are unknown. One of the most accepted hypotheses considers that both phenomena are a result of a self-limited immunologic response to pigmented cells, either in the "normal" skin or within the melanocytic lesion. Hereby we present a rare case report of a girl with halo CMN and infraorbital vitiligo. The halo CMN was excised which was followed by spontaneous improvement of vitiligo.

Key Words: *Leukoderma acquista centrifugum, Vitiligo, Excision*

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Introduction

Halo nevus, also called Sutton's nevus, is a melanocytic nevus surrounded by a halo of depigmentation, which is usually symmetrically round or oval. Halo nevi affect up to 5% children in the age group of six to fifteen years old.[1,2] in an equal sex distribution. The most common location is the back. HN has been shown to be associated with many autoimmune diseases, of which vitiligo is the most closely related with reported incidence of between 1-48%.[3,4] There are limited reports in the literature, especially with regard to CMN excision.

Case Report

A 11-year-old girl presented to the Dermatology Outpatient department for a congenital

melanocytic nevus (CMN). The lesion had been present since birth and a hypopigmented halo developed around the congenital nevus 3 years ago. She also developed hypopigmented patches developed over left periorbital region, approximately 13 months prior. Her past history and family history were unremarkable. On physical examination, the congenital nevus, measuring 0.5 cm diameter was located on the left side of nape of neck. The nevus was dark brown in colour with a depigmented halo. Examination of other body parts revealed a linear depigmented patch over the left infraorbital area. The clinical diagnosis of halo congenital melanocytic nevus associated with vitiligo was established. **(Figure 1)** The patient's parents were concerned about the appearance of the halo and vitiligo and requested to remove the CMN. Excision of the nevus with punch biopsy was performed. Pathologic examination revealed compound melanocytic nevus with congenital features and minimal lymphocytes and no macrophage infiltration, completely excised. Patient was followed up after a week to reveal a healing wound over the excision site and followed up monthly. At 5 month follow-up, the patient's halo nevus site was in a resolution phase and the vitiligo in the left infraorbital region was noted to be spontaneously repigmented **(Figure 2 & 3).**

Figure 1. Pre-excision: Halo CMN of 0.5cm in diameter with left infraorbital vitiligo

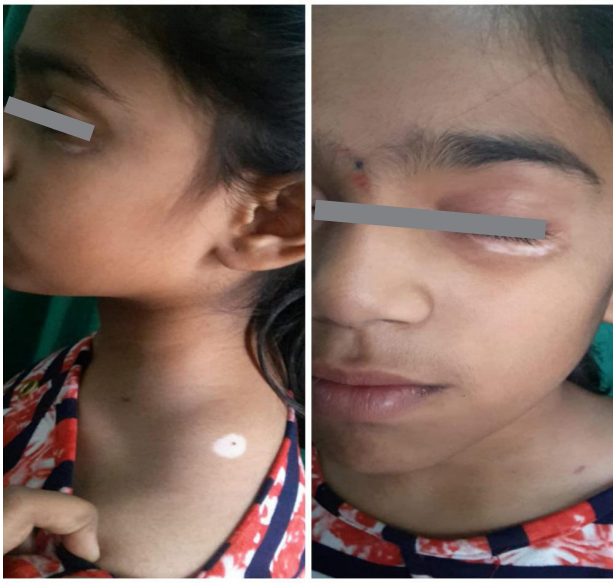


Figure 2. Gradual reduction in the size of nevi post excision in monthly visits

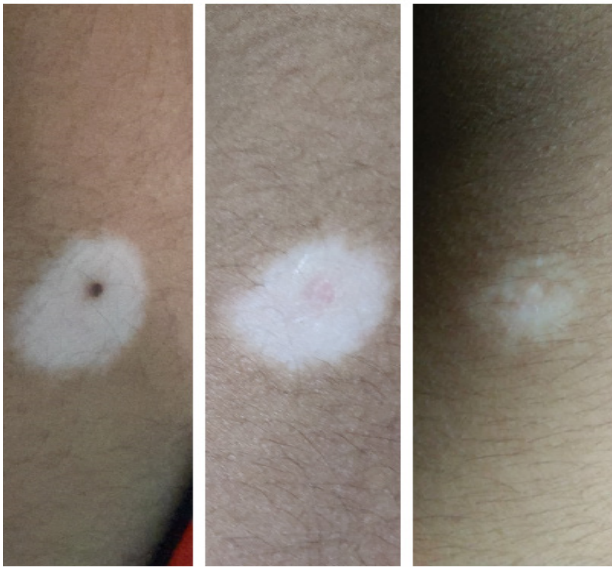
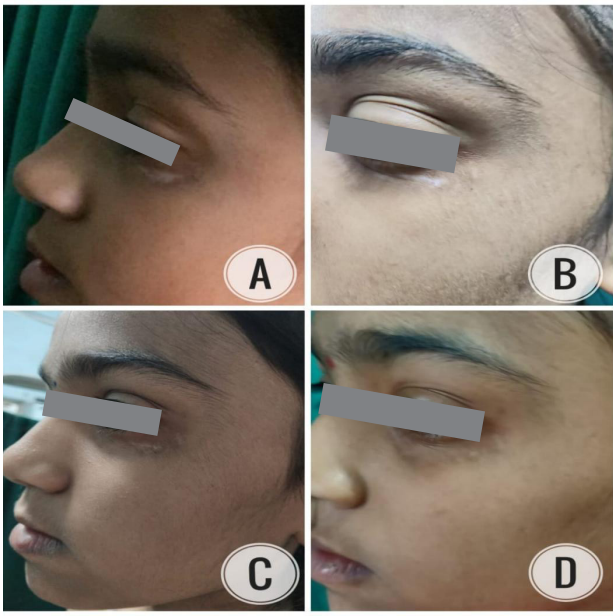


Figure 3. Spontaneous resolution of vitiligo in the left infraorbital area in monthly visits



Discussion

Halo nevi with vitiligo is well described,[5] but halo formation around congenital nevi is much less common.[6] It has been thought that similar autoimmune mechanisms, in which cytotoxic T-lymphocytes and antibodies to melanocytes (specifically IgM) cause depigmentation of the affected skin, cause vitiligo and halo nevi.[6,7] CMN with halo or vitiligo needs to be monitored clinically. The natural history of the lesion varies, because the CMN may remain

stable or undergo complete regression.[8] The risk of malignant transformation of CMN is reported to be between 1% and 5%, depending on size.[10] Surgical excision of CMN is indicated if the lesion is significantly irregular or its appearance is unstable. The resolution of vitiligo after excision is not an expected outcome, with only one prior report of excision of halo CMN with subsequent re-pigmentation of vitiligo.

In this case, excision was indicated based on the request of the patient's parents. To the best of our knowledge, there are only three prior reports of excision of halo CMN with subsequent re-pigmentation of vitiligo[8,9] in the literature. The mechanism is not quite clear. The theory of autoimmune mechanism of vitiligo and halo formation assumed that the removal of the nevus (potent "antigen") resulted in downregulation of melanocytic antibodies, leading to gradual re-pigmentation of the patient's vitiligo without further therapy. According to prior reports, re-pigmentation of the halo area often takes place over months or years, however, it does not always occur. Workman et al reported an incredibly similar case to ours, but they observed reoccurrence of depigmentation around the buttock scar 18 months after surgery. The reported reoccurrence was thought to be due to undetected residual melanocytes or antigen-presenting cells outside the margin of resection.[10]

Conclusion

In our case the rapid recovery could be due to the small size of nevus and vitiliginous area. The outcome was promising. It suggests that when antigens are removed and antibodies are cleaned out of the circulation, vitiligo may be resolved. Removing the inciting agent may be a promising way to control vitiligo.

Conflict of Interest Declaration

The authors have no conflict of interest.

Acknowledgement

Nil

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