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Pigmented colloid milium associated with exogenous ochronosis in a farmer with long-term exposure to fertilizers

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Abstract

Background: Colloid milium is a rare cutaneous deposition disorder characterized by translucent papules developing on sun-exposed regions such as the face, neck and dorsal aspects of the hands and back. Exogenous ochronosis is caused by the accumulation of homogentisic acid resulting from long-term application of certain topical agents. Histology is characterized by yellow-brown pigment deposits in the papillary dermis. Prolonged use of hydroquinone may result in the development of the pigmented form of colloid milium, sometimes in association with ochronosis.

Case report: A 53-year-old man presented with a 3-year history of multiple slow spreading pigmented papules on the dorsa of his hands, nose and ears. The patient had a long history of exposure to sun and fertilizers with no history of using hydroquinone bleaching creams. A later biopsy revealed the diagnosis of pigmented colloid milium associated with exogenous ochronosis.

Conclusion: UV light damage and long contact with fertilizers may have a role in the development of pigmented colloid milium associated with exogenous ochronosis. (*J Dermatol Case Rep*. 2015; 9(2): 42-45)

Introduction

Colloid milium is a rare cutaneous deposition disorder (deposition of amorphous material in the dermis)[1] which is characterized by translucent papules developing on light sun-exposed regions including the face, neck and dorsal aspects of the hands and back.[1,2] The four variants are (1) an adult-onset type, (2) a juvenile form, (3) a nodular form (nodular colloid degeneration), and (4) a pigmented form.[3,4] The etiology of colloid milium is uncertain. Several factors have been suggested including chronic exposure to UV light, petroleum products, hydroquinone bleaching creams and a genetic predisposition.[2] Prolonged use of hydroquinone has resulted in the development of the pigmented form of colloid milium (PCM), sometimes in association with ochronosis.[4] PCM is associated with colloid milium production resembling caviar-like papules darker than the patient's normal skin. Among the milia, atrophy may be present. The distribution is similar to that of uncomplicated exogenous ochronosis in sun-exposed areas of the face. The cause remains unknown.[1]

Exogenous ochronosis (EO) is an uncommon disorder characterized by the deposition of microscopic, ochre-colored pigment in the dermis, giving rise to a blue-black hue in the skin. It is usually manifested by asymptomatic blue-black macules on photo exposed areas, predominantly on bony prominences (malar areas, temples, lower cheeks, and neck), and it is often associated with the prolonged application of various topical chemical substances, such as hydroquinone, phenol, resorcinol, mercurial, and picric acid, as well as with quinine injections and oral antimalarial agents. Hyperpigmentation occurs strictly on the topically treated areas.[5]

We describe a case of adult pigmented colloid milium associated with exogenous ochronosis with long-term exposure to sun and fertilizers without any use of hydroquinone creams which is a very rare condition.

Case Report

A 53-year-old man presented with a 3-year history of multiple slow spreading pigmented papules on the dorsa of his hands, nose and ears came to our center. He is farmer and has a history of long exposure to sun and fertilizers and denies using any kind of hydroquinone bleaching creams or any other cosmetics as well as sun protective agents. He had no known family history of colloid milium. On examination, there were clusters of multiple pigmented (black-bluish) papules which were 5 mm in diameter on the dorsa of his hands (especially around his thumbs), nose and ears; black-bluish pigmentations under his 3rd and 4th finger nails of right hand and 1st and 2nd finger nails of his left hand (Fig. 1A, 1B).

A biopsy was performed on one of the papules of his hands. Histopathologic examination revealed nodular aggregates of amorphous cleft materials through the reticular dermis separated from epidermis by Grenz zone (Fig. 2A). The fibroblasts are aligned with the lines of fissuring colloid accompanied with melanin incontinency. Along with these are swollen yellow-brown (ochre) curled fibers irregularly placed between collagen bundles of deep dermis (Fig. 2B). The Periodic acid Schiff (PAS) and Methylen blue (MB) staining revealed positive reactivity of colloid materials (Fig. 3A, 3B). Elastin staining showed clumped elastin fibers in Grenz zone and among colloid materials. Congo red staining failed to reveal birefringence of colloid aggregates.

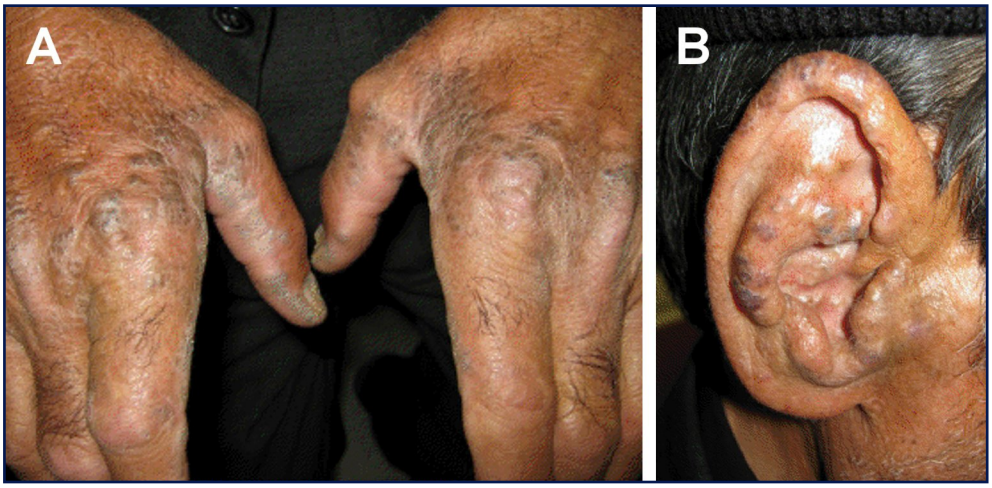

Figure 1
Black-bluish papules on the dorsa of his hands (A) and ears (B).

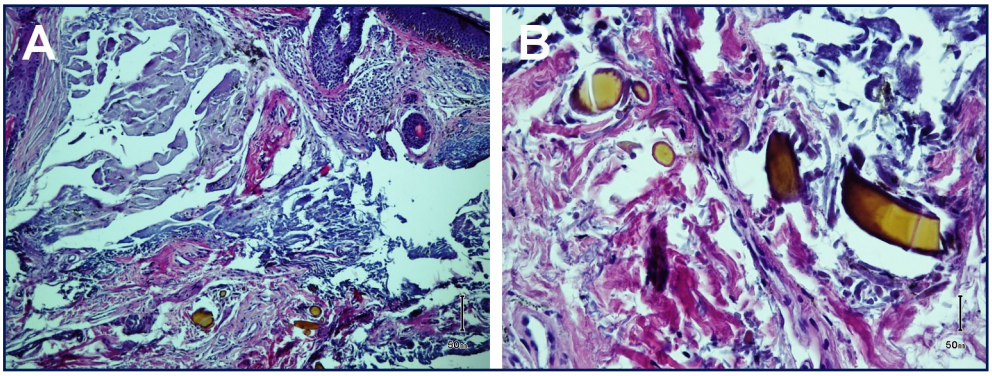

Figure 2
A. Basophilic amorphous nodules with yellow-brown fibers in deep dermis (Hematoxylin-eosin stain; original magnification ×10), B. Yellow-brown curled banana-shaped ochronotic fibers between collagen and elastin bundles (Hematoxylin-eosin stain; original magnification ×40).

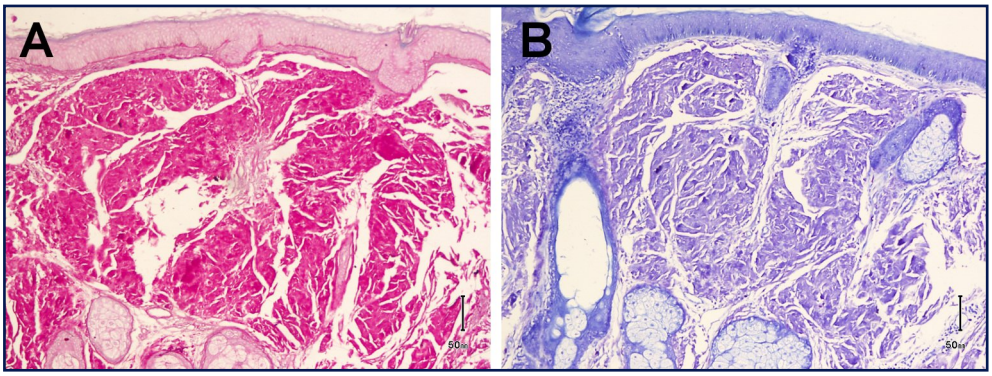

Figure 3
A. Red stained colloid materials (PAS stain; original magnification ×10), B. Blue stained colloid materials (Methylen blue; original magnification ×10).

Discussion

Colloid milia were first described by Wagner in 1866. They are characterized clinically by yellowish or gray-white translucent cysts or nodules 1 to 2 mm in diameter that develop slowly and symmetrically, reaching their maximum size in 3 years. Milia represent a degenerative change in the skin exposed to sunlight. The most commonly involved sites are the face, periorbital region, backs of the hands, back and sides of the neck, and ears. The lesions are usually asymptomatic.[6] These small, discrete, translucent cysts are firm to the touch and may release their gelatinous contents when punctured. Stroking may induce purpura in the lesions of colloid milia. No associated systemic abnormality is usually present. However, a case associated with multiple myeloma is on record. Histological examination is required to confirm the diagnosis of milia and is characterized by the presence of fissured masses of eosinophilic colloid in the papillary dermis.[6]

First reported in 1906 by Pick, ochronosis is the bluish black discoloration of certain tissues, such as ear cartilage and the ocular tissue, seen with alkaptonuria, a metabolic disorder.[7] Exogenous ochronosis (EO) is an irreversible pigmentary disorder caused by the long-term application of skin-lightening creams, most commonly containing hydroquinone.[8-12] Additionally, EO can occasionally occur from exposure to various substances such as phenol, trinitrophenol, resorcinol, mercury, picric acid, benzene[13] and antimalarial agents.[14] In the case of hydroquinone, EO may be associated with PCM,[3] which the majority of cases were in black persons. This may be because of more usage of hydroquinone bleaching creams among black people. Our case was Caucasian and he had not used any cosmetics or topical medication. He had a long history of sun exposure and long contact with fertilizers.

Without usage of hydroquinone bleaching creams their company is extremely rare.[15] To our knowledge, there is only one case of such association in published literature who was a 50-year-old white woman with esophageal squamous cell carcinoma for 3.5 years and PCM associated with EO for 4 years; she had no history of applying any hydroquinone creams; she was farmer and was exposed to excessive sunlight and chemical fertilizers just the same as our case. It was mentioned that the presence of her esophageal carcinoma with the skin EO and PCM lesions may be coincidental because her complaints of esophageal carcinoma had begun 3.5 years after the appearance of her skin lesions.

Histopathologic examination of EO reveals yellow-brown curled, banana-shaped fibers of varying thickness in upper dermis which is pathognomonic for EO. Degeneration of the ochronotic fibers finally leads to form colloid milium in advanced stages. Transepidermal elimination of pigment fibers,[16] granulomatous response,[17] pigment incontinence and solar elastosis have also been described. The major histologic differential diagnosis is lichen amyloidosis. The deposits in ACM are much larger and associated with solar elastosis and epidermal atrophy, rather than hyperplasia as seen in lichen amyloidosis.

Treatment of exogenous ochronosis remains difficult. Various treatments have been used for exogenous ochronosis such as avoidance of offending agent, retinoic acid, sunscreen, trichloroacetic acid, low-potency corticosteroids, dermabrasion, CO2 laser, cryotherapy, Q-switched ruby laser and tetracycline among others. However, the results are not satisfactory.[18]

Conclusion

We present a case of pigmented colloid milium associated with EO. Based on the observation in our patient and the last mentioned case we think that UV light damage and long contact with fertilizers may have a role in the development of this disease.

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