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# EXOGENOUS OCHRONOSIS- A CASE REPORT

**Pathology**

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## ABSTRACT

Ochronosis is rare disease portraying speckled and dispersed hyper pigmentation symmetrically over face, neck and areas exposed to sunlight. There are 2 types: Endogenous ochronosis with alkaptonuria and Exogenous which is localized and commonly caused by topical application of hydroquinine cream. Exogenous ochronosis typically presents with characteristic blue-black hyper pigmentation of skin clinching clinical diagnosis. We reported Exogenous Ochronosis in 44 year old female with history of topical application of bleaching agents for prolonged duration.

## KEYWORDS

Ochronosis, alkaptonuria.

## INTRODUCTION

The term Ochronosis is imitative of word "Ochre" in Greek, attributing to yellow discolouration.[1,2] It was described by Virchow in 1866 as deposition of brown yellow pigment in connective tissue of different organs[1,12] . It is infrequent disorder with features of speckled and diffuse pigmentation over face, neck and photo-exposed areas present symmetrically[3]. Histologically, banana shaped ochre-coloured deposits identified in dermis[3].

### Case Report

A 44 year old female presented with dark pigmentation symmetrically over malar region, nose and hands. She developed asymptomatic brown coloured pigmentation over cheeks 22 years ago. The pigment was stable. On detailed history, it was identified that she used skion lightening bleaching products on her own without any prescription. This product was used once or twice in a month over period of 10 years. This caused further involvement of nose and even hands bilaterally. The degree of pigmentation increased over period of time.

On examination, there were multiple well-defined hyper pigmented, hyperkeratotic papules coleasing to form plaques on bilateral malar, nose and bilateral hands. Remaining skin examination was unremarkable. Skin biopsy was taken from right hand and send for histo pathological examination. On Haematoxylin and Eosin stained sections, yellow-brown or ochre coloured ochronotic pigments were noted as fine granules in dermis, endothelial cells of blood vessels and also in scattered macrophages. Most important characteristic feature is ochronotic pigment seen in collagen bundles causing its swelling and homogenization. Some collagen bundles even break transversely causing homogenous, irregular, light brown clumps to lie free in tissues[3].

Diagnosis of exogenous ochronosis was made attributing to prolonged topical use of bleaching products.

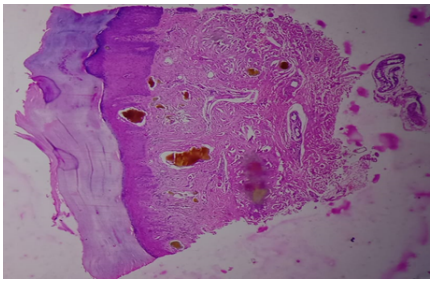

**Figure 1 : Ochre coloured deposits in dermis with homogenization of collagen bundles. (10x low power view)**

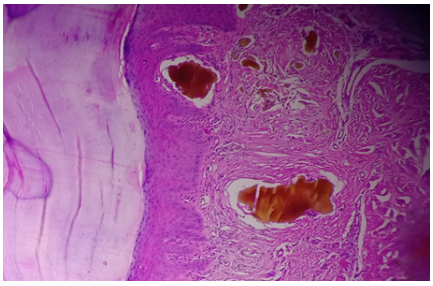

**Figure2:Yellow-brown or ochre coloured pigment in dermis and endothelial cells of blood vessels. Swelling and homogenization of collagen bundles. (40x high power view)**

## DISCUSSION

Ochronosis is unusual disorder represented by banana shaped ochre-coloured deposits in dermis. It is of two types- exogenous and endogenous ochronosis.

Pick described Exogenous Ochronosis in 1906[1,14]. Beddard and Plumtre in 1912, described the disease when phenol was used for leg ulcer by patient[1,13]. De Beer illustrated Exogenous Ochronosis in patients using hydroquinone as the major constituent of topical lightening agents[1,15].

Exogenous ochronosis is clinically and histologically identical to alkaptonuria[1]. Manifested as bilaterally symmetrical,asymptomatic blue-black speckled macules. It was interpreted to be "caviar like bodies" involving malar temple areas, neck and lower cheeks. Usually due to topical use of bleaching cream containing hydroquinone. It is also related to use of resorcinol, phenol, picroc acid, quinine injections and oral anti-malarials.

Endogenous ochronosis, also named as "Black Urine Disease" or "Alkaptonuria", is inherited autosomal recessive disorder due to defect in chromosome 4q23. This leads to inactivation of homogentisate 1,2 dioxygenase, further causing accumulation of homogentisic acid in liver. It is water soluble and polymerised to brownish-black pigments aggregated in connective tissue. It binds irreversibly to dermal fibrillar collagen causing pigmentation of skin. Musculoskeletal instances are seen considerably in this cases.[1]

According to Penneys theorey[16], hydroquinone causes inhibition of enzyme homogentisic oxidase causing aggregation of homogentisic acid along with its metabolic products. This further polymerizes forming ochronotic pigment in papillary dermis.

Histopathology remains gold standard, although invasive, for diagnosis of Exogenous Ochronosis. Characteristic feature is ochre-coloured banana shaped fibres in dermis. Also seen is in reticular and papillary dermis along with swelling and homogenization of collagen bundles. For skin biopsy to be more accurate, speckled macular lesions are preferred.

In our case, rather than being high concentration of the chemical agent, it was prolonged use of bleaching products without any knowledge.

## CONCLUSION

To recapitulate, Exogenous Ochronosis is an underdiagnosed entity. Its diagnosis is usually missed clinically. Hydroquinone containing topical depigmenting agents, when used for longer duration is prevalent cause of Exogenous Ochronosis. Early diagnosis is crucial so that adequate measures are taken by preventing usuage of hydroquinone.

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