Eruptive segmental collagenoma of the face in a young child: A rare case report

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ABSTRACT

Collagenomas are benign connective tissue dermal hamartomas of unknown etiology, characterized by an imbalance in the distribution and amount of collagen in the extracellular matrix. Very few cases are reported in children as most cases are seen in young adults. Case of a five years old girl is described, who presented with asymptomatic, skin colored infiltrated papules and plaques on the face with no systemic involvement diagnosed as eruptive collagenoma.

Collagenoma, Hamartoma, Child, Face

INTRODUCTION

Connective tissue nevi of the skin are benign dermal hamartomas that consist predominantly of one of the components of extracellular matrix like collagen, elastin or proteoglycans. Connective tissue nevi that are predominantly composed of collagen are called collagenomas.¹ Collagenomas are most commonly seen in the first two decades of life and are usually reported in adult life, with lesions predominantly seen over the extremities.1 histopathological and On examination, the lesions show an increased amount of collagen fibers and decreased fragmented elastic fibers in the dermis. Thickened and haphazardly arranged dense fascicular bundles of collagen are seen with Masson's trichrome stain.²

CASE REPORT

A mother brought her five-year-old daughter to skin OPD in November 2017 with complaint of otherwise asymptomatic, yellowish looking lesions on left cheek and right upper eyelid, which started to appear at around six months after her birth and were gradually increasing. (Figure 1). The lesions appeared only on face without involving trunk or extremities and no history of any preceding inflammation or trauma was narrated. There was no history of epilepsy, congenital anomalies or delayed milestones, and other siblings were not having similar or any other skin problem. On examination, multiple well-defined, cutaneous infiltrated, grouped, painless, skin-colored to yellowish papules and plagues with a leathery surface were present

in a dermatomal distribution, without crossing the midline over the right upper eyelid and left cheek. According to the parents, the lesions were also increasing gradually and becoming more prominent as the child was growing in age. No lesions were identified on any other body site and systemic examination was completely normal. ΑII baseline laboratory investigations were normal. Differential diagnosis included connective tissue nevi, nevus mucinosis, mastocytoma and juvenile xanthogranulomas. A 2 mm punch biopsy was obtained from the lesion present on cheek. On histopathology, epidermis was intact while papillary and reticular dermis showed increased proliferation of collagen fibres, suggestive of collagenoma. Special stains for elastic fibres were requested but could not be performed due to nonavailability in the institutional histopathology laboratory and non-affordability by patient from an outside laboratory. Final diagnosis of `Eruptive Segmental Collagenoma' was made. Treatment was planned to carry out fortnightly sessions of electrocauterization with plasma pen after topical application of lignocaine gel, but the child was not very cooperative.1 It was decided to defer the treatment till the patient is mature enough. The parents were reassured of the benign nature of the condition and counseled to have regular follow ups every six months. There was no progression of existing lesions or any formation of new lesions seen at subsequent follow up for 18 months.

DISCUSSION

Collagenomas are hamartomas and classified as localized or generalized and inherited or acquired.

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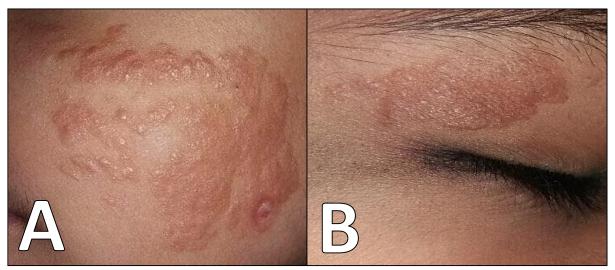


Figure 1. A) Left cheek showing lesion. B) Right upper eyelid showing lesion

Hereditary autosomal dominant collagenomas include Shagreen's patches of Tuberous Sclerosis and familial cutaneous collagenomas, and acquired ones include eruptive collagenomas and isolated collagenomas.²⁻⁶ Isolated collagenomas are sporadically acquired collagenomas that are localized to single region and are not associated with any disease. Isolated collagenomas can present as plantar collagenoma, zosteriform lesions and papulo-linear lesions.8 Familial collagenomas are inherited in an autosomal dominant fashion and includes cutaneous collagenomas associated with cardiovascular abnormalities.9 Present patient was categorized as acquired segmental variety of eruptive segmental collagenoma on the basis of first decade of presentation, multiple lesions typical of collagenoma, involving one segment of right upper eyelid and left maxillary region, biopsy findings, absence of family history and no other systemic involvement.

Though in most cases the onset of presentation is beyond the first decade of life, this patient presented with onset of lesions soon after birth. The earliest onset of presentation reported in the published indexed literature was found to be in a 2-year old Nigerian girl and another with onset of lesions at 5 years of age. 10,11 Two previous reports could be traced to describe predominant involvement of the face in addition to other more common sites. 10,12 The lesions are benign so no active intervention is required. One previous study demonstrated therapeutic effect of intralesional triamcinolone acetonide on cutaneous collagenomas, with improvement of the lesions in varying degrees. 13 Due to the large size of the lesion and poor cosmetic

outcome of the surgical excision, it was decided to go for electro-cauterization. However, treatment was deferred as child was not cooperative. Eruptive Collagenoma of the face is a relatively uncommon disorder with very few case reports in pediatric population. To the best of authors' knowledge, this is the first case of facial variant of 'Eruptive Segmental Collagenoma' in pediatric age group to be published from Pakistan.

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