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Isolated collagenoma on the face: a rare occurrence

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Abstract

Collagenomas are connective tissue nevi with hamartomatous proliferations of dominant dermal collagen. They can present as solitary or multiple inherited or acquired lesions over various body sites. The face is a rare site of collagenomas and, of the few cases reported in the literature, they have been seen more often on the scalp or on the plantar area. An extensive literature search did not reveal any cases of isolated collagenoma on the face. Herein we present the case of 22-year-old female with isolated collagenoma on the face. This case is being reported because of its unique location and rarity.

Keywords: collagenoma, isolated, nevi

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Introduction

Connective tissue nevi are hamartomatous proliferations of the connective tissue components in the dermis. Uitto et al. classified them on the basis of their inheritance pattern and the involvement of the predominant extracellular connective tissue component: collagen, elastic fibers, or proteoglycans (1). Those with dominant dermal collagen are identified as collagenomas and have been specified as the Lipschutz type, whereas those with predominant elastic tissue changes are the Lewandowsky type (1). They can be solitary or multiple, and inherited or acquired. Collagenomas usually present as asymptomatic skin-colored plaques, papules, or nodules of various sizes in solitary, grouped, linear, or irregular distribution. Acquired collagenomas are called eruptive collagenomas when multiple, and when they are single or restricted to one body site they are called isolated collagenomas (2). Collagenomas usually occur over the upper trunk, arms, back, thighs, and soles (2). The face is a rare site of collagenomas and, among the few cases reported in the literature, they have been more often seen on the scalp or on the plantar area. To the best of our knowledge, only 18 reported cases of isolated collagenoma could be retrieved in English literature by searching on PubMed, out of which the majority of cases were on the scalp, back, and palmo-plantar areas. One case each was seen on the frontal area and labia majus (Table 1). Comprehensive search of the literature yielded no descriptions of isolated collagenoma on the face to date.

Case report

A 22-year-old female presented to the dermatology department with eruption of multiple, coalescing skin-colored to brown papules varying from 2 mm to 10 mm in size on the left side of the face that had appeared 6 months prior (Fig. 1). The lesions were asymptomatic but progressive. The history of injury and family history of similar skin lesions was negative. Systemic examination and routine hematological and biochemical investigations were within normal limits. Clinical differentials of connective tissue nevus, nevus sebaceous, and nevus lipomatosis superficialis were made. A punch biopsy was taken, which upon histopathological examination showed mildly acanthotic epidermis with mild nonspecific chronic perivascular inflammation in the upper dermis. Lobules of acellular, collagenized connective tissue in a haphazard arrangement were seen in the reticular dermis (Fig. 2) insinuating between the dermal appendages. The adnexal structures were preserved and no inflammatory cells were observed around them. Verhoeff–Van Gieson (VVG) stain for elastic fibers revealed marked reduction and fragmentation of elastic fibers (Fig. 3), thereby confirming that the connective tissue lobules consisted of collagen. Based on these findings, the patient was diagnosed as having isolated collagenoma. No specific treatment was given because the patient was lost to follow-up.

Discussion

Collagenomas are now usually classified into four groups, two inherited and two acquired. The inherited ones are 1) familial cutaneous collagenomas and 2) shagreen patches of tuberous sclerosis, and the acquired ones are 3) eruptive collagenomas and 4) isolated collagenomas. Familial cutaneous collagenomas have an autosomal dominant inheritance, are present in a symmetrical distribution on the upper trunk, and are associated with extracutaneous abnormalities such as sensorineural hearing loss, recurrent vasculitis, and cardiac disorders such as idiopathic cardiomyopathy

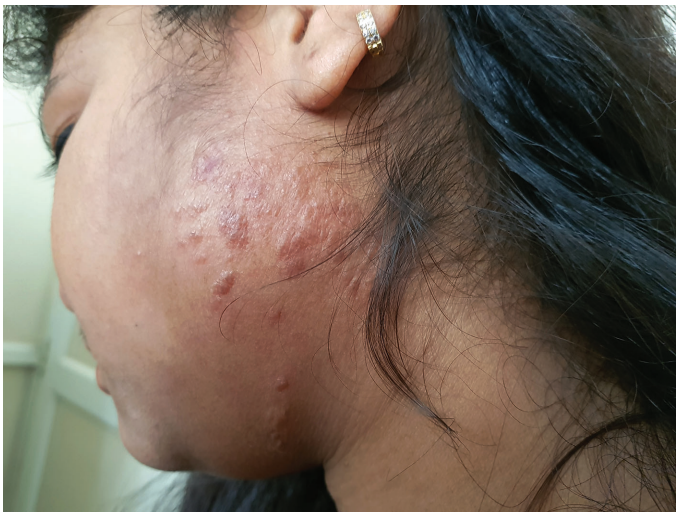

Figure 1 | Multiple skin-colored papules varying from 2 mm to 10 mm in size on the left side of face.

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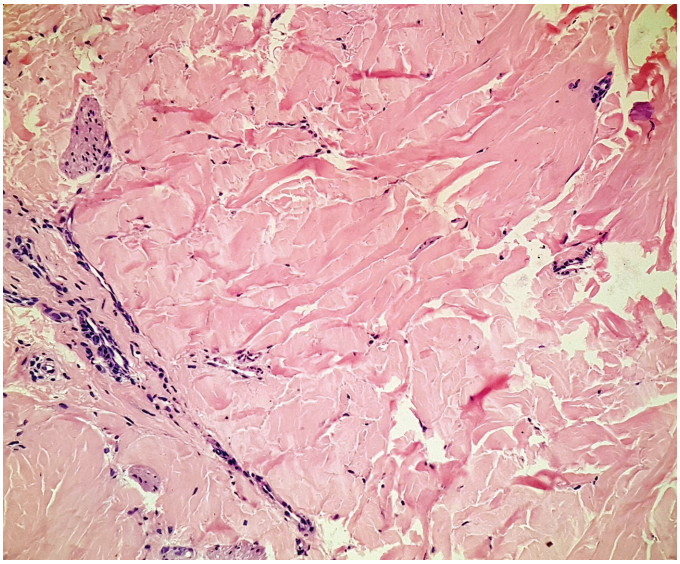

Figure 2 | Acellular, thickened, and condensed collagen in a haphazard arrangement in the reticular dermis (H&E, 200×).

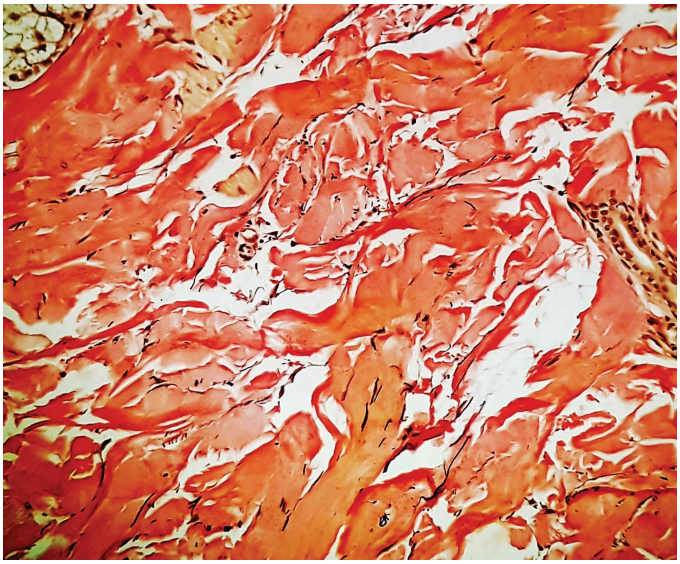

Figure 3 | Fragmented, curled, and denatured elastic fibers in the dermis (VVG, 400×).

Table 1 | Eighteen cases of isolated collagenoma.

No.	Country	Age (years)	Sex	Family history	Site	Histopathology	Treatment	Workup	References
1	Taiwan	8	M	–	Left upper arm	Papulolinear collagenoma	Not mentioned	Not mentioned	3
2	Brazil	3	F	–	Lower back	Papulolinear collagenoma	Not mentioned	MRI, CT scan	6
3	Brazil	45	F	–	Hands, fingers	Papulolinear collagenoma	Not mentioned	Skeletal survey	7
4	Turkey	26	F	–	Proximal part of left arm	Eruptive collagenoma	Refused treatment	Not mentioned	8
5	Iran	14	M	–	Frontal	Isolated collagenoma	Intralesional triamcinolone acetonide injections	MRI, CT scan	9
6	India	23	F	–	Labium majus	Isolated collagenoma	Refused treatment	Not mentioned	10
7	India	20	M	–	Scalp	Isolated collagenoma	Surgery	MRI, CT scan	2
8	India	18	F	–	Lower back	Isolated corymbose collagenoma	Intralesional triamcinolone hyaluronidase	MRI, CT scan	11
9	India	20	M	–	Scalp	Isolated pedunculated collagenoma	Surgical excision	MRI, CT scan	4
10	India	9	F	–	Plantar of right foot	Isolated cerebriform collagenoma	Not mentioned	Skeletal survey	12
11	Japan	6	F	–	Scalp	Isolated collagenoma	Not mentioned	Not mentioned	13
12	United States	40	F	–	Toe, plantar of right foot	Isolated collagenoma	Not mentioned	Not mentioned	14
13	India	35	F	–	Scalp	Isolated collagenoma	Not mentioned	Skeletal survey	15
14	Turkey	19	F	–	Plantar right foot	Isolated collagenoma	Surgical excision	ECG, echo skeletal survey	16
15	Korea	23	F	–	Toe, plantar of right foot	Isolated collagenoma	Not mentioned	Not mentioned	17
16	India	22	M	–	Palm	Isolated collagenoma	Not mentioned	Not mentioned	18
17	Switzerland	11	M	–	Plantar of right foot	Isolated collagenoma	Surgeon refused excision	Skeletal survey	19
18	Spain	6	F	–	Toe, plantar of right foot	Isolated collagenoma	Not mentioned	Not mentioned	20
19	India	22	F	–	Face	Isolated collagenoma	No follow-up	–	Present case

F = female, M = male, MRI = magnetic resonance imaging, CT = computed tomography.

and congestive heart failure. Shagreen patches of tuberous sclerosis also have a family history and are associated with other characteristic cutaneous manifestations of adenoma sebaceum, subungual fibroma, and ash leaf macules (2). Both eruptive and isolated collagenomas are acquired connective tissue nevi of the collagen type and lack a family history. Although the histopathological features of both are the same, isolated collagenomas are localized to a single body region, as in our case. Varying presentations of isolated collagenomas such as paving stone nevi, plantar fibromatosis, and papulolinear and zosteriform lesions have been reported in the literature (3, 4). The pathogenesis of collagenomas or connective tissue nevi is unclear and, because they are benign, no specific treatment is currently given in most cases (2). According to Uitto et al., collagenomas are composed exclusively of type I collagen. They form due to reduced production of collagenase, causing reduced degradation of collagen locally (5). Histopathological examination is the gold standard for diagnosis. In diagnosed cases, a further workup is necessary to rule out any underlying systemic disorder because collagenomas have been associated with disorders such as hypogonadism, pseudohypoparathyroidism, and Down syndrome (2). Diagnosis of isolated collagenoma was made by a combination of standards, including lack of family history and extracutaneous manifestations, single location, and classical clinical and histopathological features.

We are reporting this case because of its unique location, rarity, and absence of any associated abnormalities. Publication of such cases should be encouraged because this may be an underdiagnosed entity and awareness of it would improve the recognition of this condition.

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