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Unravelling a Rare Dermatologic Entity: Case Report on Storiform Collagenoma

Dr. Naqsh Thakur[1], Dr. Yogita Bhansali[2], Dr. Tapsh Thakur[3]
[1,2] Department of Pathology, Shri B.M Patil Medical College, Hospital and Research Centre, Vijayapura, Karnataka
[3] Department of Surgery, Dr. Rajendra Prasad Govt. Medical College, Kangra, Himachal Pradesh
Corresponding Author: *Dr. Yogita Bhansali*
Email: *yogitabhansali2@gmail.com*

Abstract:
Storiform collagenoma, also known as sclerotic fibroma, is a rare and benign skin tumor characterized by the proliferation of fibroblasts and excessive production of type I collagen. These tumors most commonly present as painless, slow-growing, well-circumscribed nodules on the face, neck, and extremities, though they can occur in other locations. While usually solitary, multiple lesions may indicate Cowden syndrome, a genetic condition associated with increased cancer risk. This report presents the case of a 42-year-old woman with a storiform collagenoma on her left forearm. The tumor was surgically excised, with no recurrence at a 12-month follow-up. This report underscores the importance of accurate diagnosis and proper management for favourable outcomes.
Keywords: Collagenoma, Fibroma, Storiform pattern, Cowden syndrome.

Introduction

Storiform collagenoma, also known as sclerotic fibroma, is a rare type of benign cutaneous tumor characterized by the proliferation of fibroblasts and the formation of thick collagen bundles arranged in a storiform pattern (Rapini & Golitz, 1989). This lesion primarily affects young to middle-aged adults of both sexes, although a slight female predominance has been reported (Lee et al., 2007). These tumors most often present as painless, slow-growing, well-circumscribed nodules of skin-colored or whitish appearance (Requena et al., 1992).

Storiform collagenomas can occur as solitary or multiple lesions. The solitary form is generally sporadic and not associated with any systemic condition (Donati et al., 1991). However, the presence of multiple lesions may serve as a dermatological marker for Cowden syndrome, a genodermatosis with an autosomal dominant inheritance pattern linked to mutations in the PTEN gene (Al-Daraji et al., 2007). Cowden syndrome predisposes patients to various malignancies, including breast, thyroid, and endometrial cancers (Weary et al., 1972).

The pathogenesis of storiform collagenoma remains uncertain. Some authors have classified these tumors as hamartomas, while others consider them to be true neoplasms (Shitabata et al., 1995). Histopathologically, they are composed of dense, hypocellular collagen fibers arranged in a storiform or "onion-skin" pattern, surrounded by normal dermal and epidermal structures (Izquierdo et al., 2001). Immunohistochemistry typically reveals positivity for CD34 and vimentin, supporting their fibroblastic origin (Bhambri & Del Rosso, 2009).

Accurate diagnosis of storiform collagenoma is essential to distinguish it from other benign and malignant cutaneous lesions, including dermatofibroma, pleomorphic fibroma, and

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sclerotic lipoma (Rudolph et al., 1998). Treatment usually involves complete surgical excision, which is curative for solitary lesions (Tosti et al., 1999).

Case Details

A 42-year-old woman presented with a painless, slow-growing nodule on her left forearm. The lesion had been present for approximately two years without any significant changes in size or appearance. The patient denied any history of trauma at the site, systemic symptoms, or similar lesions elsewhere on the body.

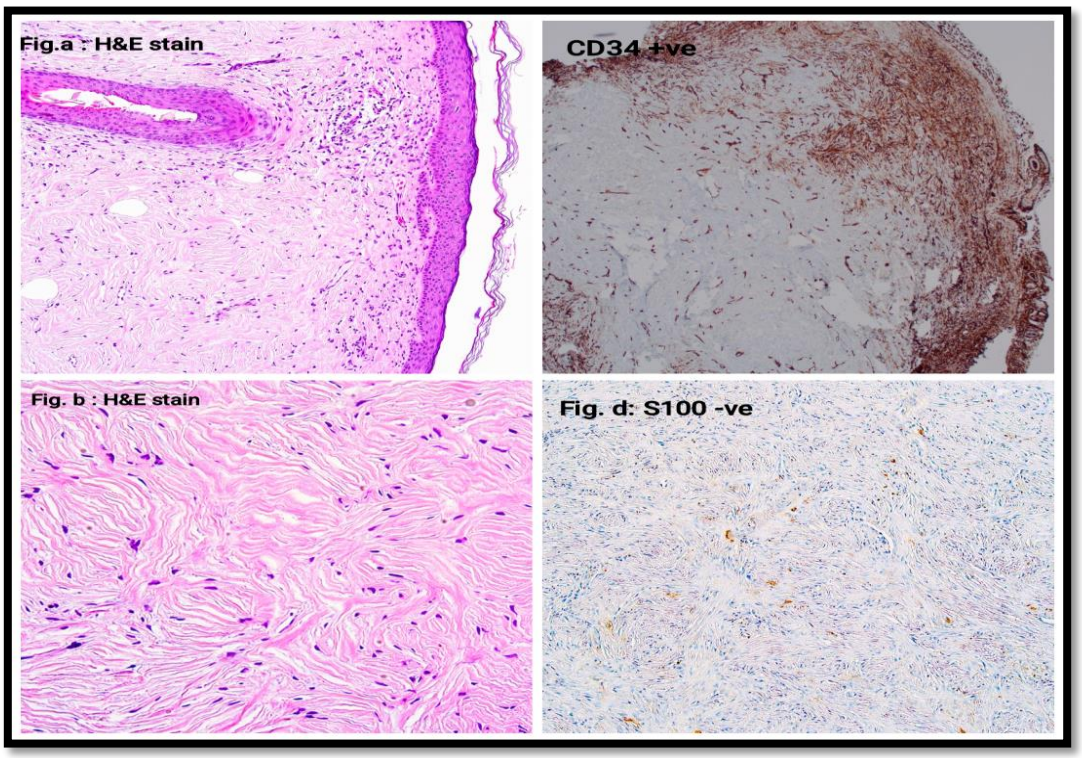
On physical examination, a well-circumscribed, firm, skin-colored nodule measuring 1.8 cm in diameter was observed on the dorsal aspect of the left forearm. The surface was smooth, and the lesion was non-tender upon palpation. No regional lymphadenopathy was noted.

The patient's medical history was unremarkable, with no personal or family history of Cowden syndrome or other genetic conditions. Routine laboratory tests, including thyroid function and metabolic panel, were within normal limits.

Given the clinical presentation, a provisional diagnosis of a benign cutaneous tumor was made. The lesion was excised under local anaesthesia, and the specimen was sent for histopathological evaluation.

Histopathological examination revealed a well-defined nodule composed of dense, hypocellular collagen bundles arranged in a storiform pattern. The surrounding dermis and epidermis appeared normal, with no evidence of malignancy. Immunohistochemical staining was positive for CD34 and vimentin but negative for S-100 and epithelial markers, confirming the diagnosis of storiform collagenoma. (Figure a, b, c, d).

The postoperative course was uneventful, with complete wound healing observed at two weeks. Follow-up at 12 months showed no signs of recurrence.



Discussion

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Storiform collagenoma is a rare dermatological entity that poses diagnostic challenges due to its resemblance to other fibrous skin tumors (Rapini & Golitz, 1989). The storiform arrangement of collagen fibers is a defining histopathological feature that aids in its identification (Requena et al., 1992).

The differential diagnosis of storiform collagenoma includes dermatofibroma, sclerotic lipoma, pleomorphic fibroma, and benign fibrous histiocytoma (Izquierdo et al., 2001). Dermatofibromas typically exhibit a more cellular and less organized collagen pattern, while sclerotic lipomas contain mature adipose tissue interspersed with fibrous components (Zelger et al., 1997). Immunohistochemistry is a valuable tool in distinguishing storiform collagenoma from these lesions, as it typically shows positivity for CD34 and vimentin but negativity for S-100 and epithelial markers (Bhambri & Del Rosso, 2009).

The association between multiple storiform collagenomas and Cowden syndrome underscores the importance of thorough clinical evaluation and genetic counseling when multiple lesions are present (Al-Daraji et al., 2007). Cowden syndrome is a genodermatosis characterized by mucocutaneous lesions, benign and malignant neoplasms, and a predisposition to breast, thyroid, and endometrial cancers (Weary et al., 1972). Early diagnosis of Cowden syndrome can facilitate cancer surveillance and improve patient outcomes (Requena et al., 1992).

The pathogenesis of storiform collagenoma remains a topic of debate. Some authors propose that it represents a hamartomatous lesion resulting from dysregulated fibroblast activity, while others consider it a true neoplasm with ongoing collagen synthesis (Shitabata et al., 1995). The "onion-skin" arrangement of collagen fibers observed in histopathological specimens supports the hypothesis of aberrant fibroblast activity (Izquierdo et al., 2001).

Treatment for storiform collagenoma typically involves complete surgical excision, which is curative for solitary lesions (Tosti et al., 1999). Recurrence is rare, and no cases of malignant transformation have been reported (Donati et al., 1991). However, long-term follow-up may be warranted in cases associated with Cowden syndrome to monitor for the development of malignancies (Al-Daraji et al., 2007).

Conclusion

Storiform collagenoma is a rare and benign cutaneous tumor that can occur as a solitary lesion or in association with Cowden syndrome. Accurate diagnosis through histopathological examination and immunohistochemistry is essential to distinguish it from other fibrous skin tumors. Surgical excision is curative for solitary lesions, with an excellent prognosis. Awareness of this rare tumor and its potential association with Cowden syndrome can facilitate early diagnosis and appropriate management.

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