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Solitary papule over scalp

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**Case presentation**

**Solitary papule over scalp**

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**Abstract**

Folliculosebaceous cystic hamartoma (FSCH) is a rare cutaneous hamartoma characterized by follicular, sebaceous, and mesenchymal elements. Folliculosebaceous cystic hamartoma is probably not as rare as previously thought and its inclusion in the differential diagnosis of asymptomatic skin colored papules or nodules is warranted, especially if it is present in the head and neck region.

**Key words: folliculosebaceous cystic hamartoma, sebaceous tumor**

**Case synopsis**

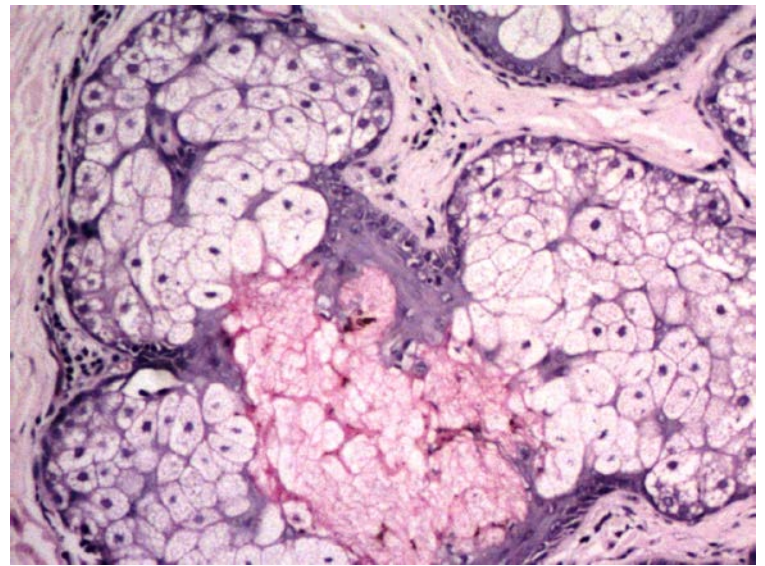
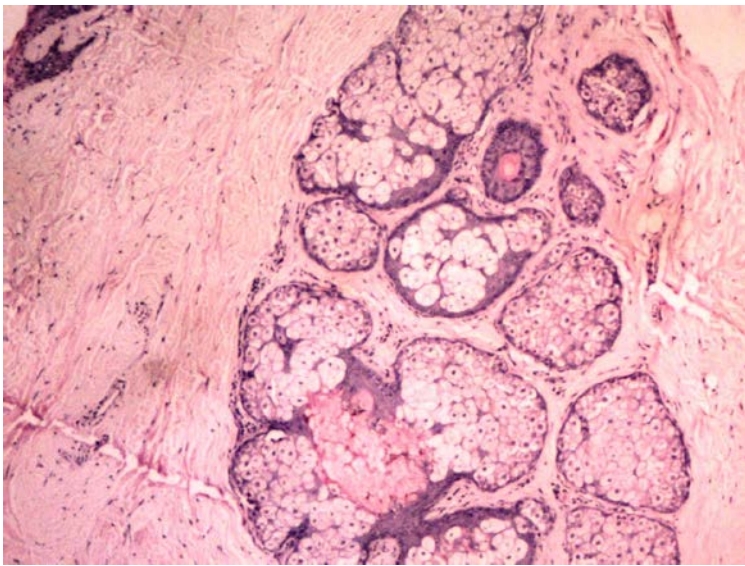
A 33-year-old woman presented with an asymptomatic papule that had persisted for the past 11 years. She noticed slow growth in the size of the lesion over the past 5 years. Repeated trauma to the papule while combing her hair resulted in discomfort.

Physical examination revealed a single non-tender, skin colored, firm, hairless papule of 5 x 4 x 3 mm diameter over the vertex of the scalp (Figure 1). It was excised and sent for histopathological examination (Figure 2, 3 & 4).

Histopathological examination revealed a dilated follicular cystic structure with numerous sebaceous lobules radiating out from it in the dermis (Figure 2). The cyst showed a predominantly infundibular keratinization (Figure 3). This folliculosebaceous structure was surrounded by increased collagen in the dermis (Figure 2) and clefts were visible between the folliculosebaceous structures and the surrounding stroma (Figure 4). The stroma consisted of collagen, few capillaries, adipocytes, and scattered lymphocytes (Figure 5-7).

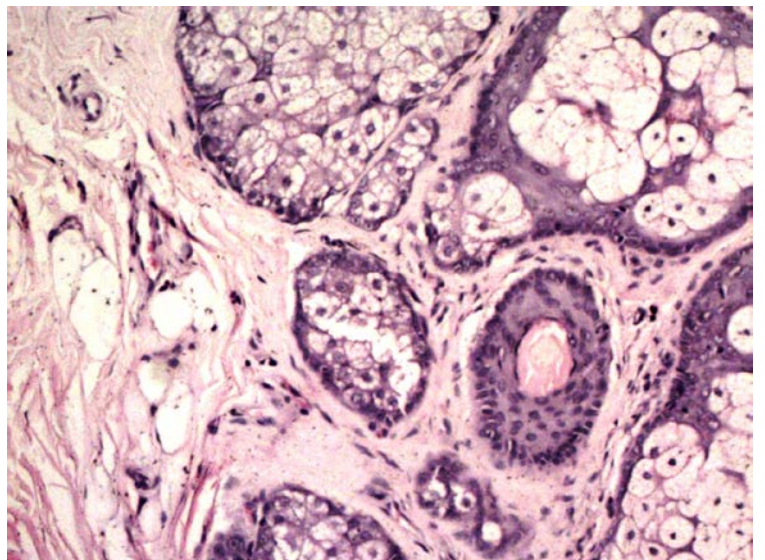
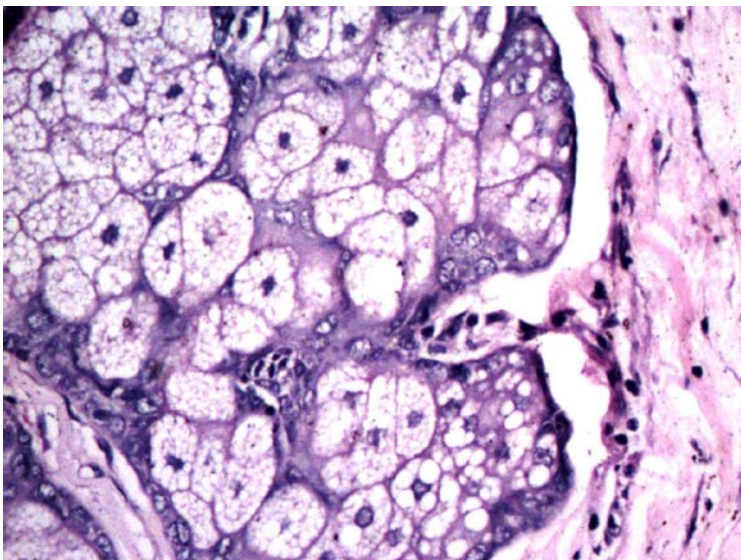


**Figure 1.** Single skin colored, firm, hairless exophytic papule over vertex of scalp



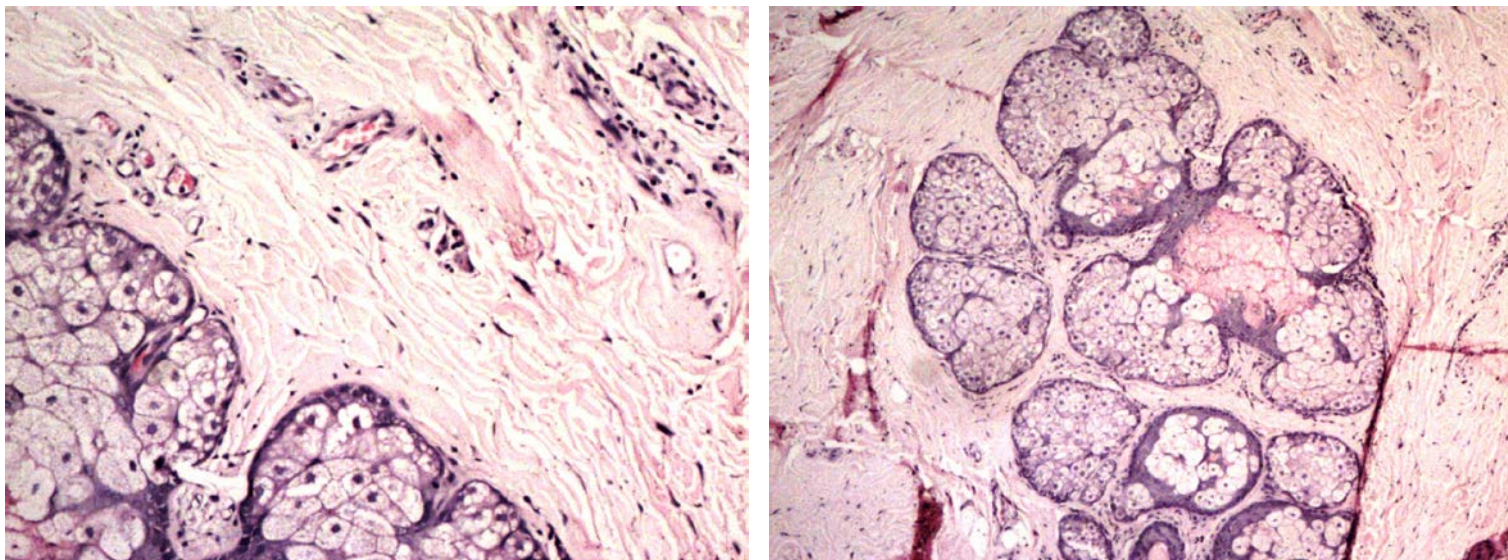
**Figure 2.** Dilated follicular cystic structure with numerous radiating sebaceous lobules in the dermis. (H&E; 40X)

**Figure 3.** Infundibular keratinization in the dilated follicle from which sebaceous lobules are seen radiating out. (H&E; 100X)



**Figure 4.** Clefts visible between the folliculosebaceous structures and surrounding stroma. (H&E; 100X)

**Figure 5.** Stroma surrounding the folliculosebaceous structure consisting of collagen bundles interspersed with few adipocytes and capillaries. (H&E; 100X)



**Figure 6.** Stroma surrounding the folliculosebaceous structures consisting of collagen bundles interspersed with multiple capillaries and sparse lymphocytes. (H&E; 100X)

**Figure 7.** Dilated follicular cystic structure with numerous radiating sebaceous lobules in dermis, surrounded by stroma consisting of collagen, capillaries, few adipocytes, and sparse scattered lymphocytes. (H&E; 40X)

## Discussion

Folliculosebaceous cystic hamartoma (FSCH) is a rare cutaneous hamartoma characterized by follicular, sebaceous, and mesenchymal elements. It was initially described by Kimura et al in 1991 [1]. Initially, this was considered a rare entity, but in the recent past quite a few cases have been reported [2].

Folliculosebaceous cystic hamartoma usually occurs on the head and neck as a papule or nodule less than 25mm in size [3]. Cases have also been reported on other areas such as the nipple [4], genitalia [5], upper limb [6], lower limb [7], and upper back [8]. Folliculosebaceous cystic hamartoma is usually seen in adults but congenital cases have been reported [5, 8]. In the reported cases of FSCH, the initial clinical diagnosis suspected varied from benign conditions like intradermal nevus, sebaceous hyperplasia, benign soft tissue neoplasms (lipoma, neurofibroma) to malignant ones like basal cell carcinoma [3, 9]. FSCH lacks distinctive clinical features and is usually unsuspected until the histopathology shows the characteristic findings. There has been no evidence of spontaneous regression in FSCH [1, 9].

On histopathology, FSCH characteristically presents as a dermal lesion, with a central infundibular cyst of the hair follicle, with sebaceous glands radiating out, and abundant stroma, which forms the mesenchymal component [1]. The mesenchymal component of FSCH is highly polymorphous containing collagen, fat, blood vessels, and neural tissue in varying proportions [10]. Clefts may be visible between fibroepithelial units and surrounding stroma. Laminated fibroplasia is usually seen around the epithelial component [1]. In our case, there was a dermal lesion consisting of a dilated follicular cystic structure with infundibular keratinization, radiating sebaceous follicles, and surrounding clefts between folliculosebaceous structures and surrounding stroma. In our case, the stroma mainly consisted of bundles of collagen with interspersed capillaries and occasional adipocytes.

The differential diagnosis of FSCH includes sebaceous trichofolliculoma (trichofolliculoma with marked sebaceous components) and fibrofolliculoma. Sebaceous trichofolliculoma clinically presents as an endophytic nodule with central umbilication containing short pigmented hair in clusters, whereas FSCH presents as an exophytic papule or nodule which may be sessile or pedunculated; no umbilication or pigmented hair is visible. Folliculosebaceous cystic hamartoma has a prominent mesenchymal component unlike sebaceous trichofolliculoma. The presence of multiple hair shafts inside the dilated follicular infundibulum is a characteristic of sebaceous trichofolliculoma but not seen in FSCH [9]. Fibrofolliculoma is a benign tumor of the perifollicular sheath, which presents as asymptomatic small multiple (rarely solitary) dome shaped papules over scalp, face, neck, and upper trunk. On histopathology, it is characterized by a central dilated hair follicle with keratinous plug, with radiating anastomosing strands of branching basaloid and angiofibromatous perifollicular stroma [11]. It can be differentiated from FSCH by the

conspicuous absence of sebaceous glands. There are reports of FSCH arising within port-wine stains [12] and FSCH associated with nevus lipomatosus [13].

The histogenesis of FSCH is not known but the immunohistochemistry reveals a profile very similar to sebaceous glands [14]. Schulz et al [15] had proposed that FSCH is not a distinct entity but a very late stage of trichofolliculoma. However, the immunohistochemistry of FSCH suggests that adipocytes and other mesenchymal components in FSCH may be originating from nestin positive pluripotent stem cells. The high nestin expression and prominent mesenchymal component of FSCH, unlike that seen in trichofolliculoma, suggests that both the conditions do not represent chronological changes of the same entity, but are rather two distinct entities [16]. Folliculosebaceous cystic hamartoma is probably not as rare as previously thought and its inclusion in differential diagnosis of asymptomatic skin colored papules or nodules is warranted, especially if it is present in the head and neck region.

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