Journal of Science and Technology, Vol. 42, No. 4 (2024), pp 11 - 16

https://dx.doi.org/10.4314/just.v42i4.2

MULTIPLE TRICHOFOLLICULOMAS IN AN ADOLESCENT FEMALE: A CASE REPORT

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ABSTRACT

Trichofolliculoma is a rare benign tumour of hair follicle tissue that is often found on the face and neck. It is usually a dome-shaped papule or nodule with a central pore and a tuft of immature hairs growing through it. It typically presents as a solitary lesion, with onset in adulthood.

We report a rare case of multiple facial trichofolliculomas in an adolescent female, with onset in childhood. The patient was a 12-year-old who presented with non-pruritic facial lesions of seven years duration that were occasionally painful. On examination, multiple skin-coloured nodules and papules with central pores confined to the face were noted. The lesions were numerous, hence presenting a challenge in individually counting them. Nevertheless, we estimated the number of lesions to exceed 120. There was no hair, nails, mucosae, or systemic involvement.

The clinical diagnosis of trichofolliculoma was confirmed with an excision biopsy of a single lesion for histopathology. Treatment was with curettage under local anaesthesia for cosmetic reasons. Healing was rapid (within four to five days) with no significant scarring or recurrence after six months of follow-up.

Multiple trichofolliculomas, though uncommon, can manifest in young patients. Histological diagnosis is critical in treatment because it helps to rule out similar conditions.

Keywords: Multiple, benign, follicle, trichofolliculomas, case report.

INTRODUCTION

Trichofolliculoma is an uncommon hamartoma of hair follicle tissue usually located on the face (frequently around the nose) and neck.(El-Komy and Abdelkader 2020) The exact incidence of the tumor has not been determined because no large screening has been performed in the general population.(El-Komy and Abdelkader 2020) It is clinically described as a dome-shaped papule with a central pore, characteristically with a wool-like wisp of rudimentary hairs growing through it.(Pinkus and Sutton 1965) The central pore usually drains sebaceous-like material.(Schulz and Hartschuh 1998) Trichofolliculoma usually occurs in middle-aged adults (mean age of 45 years) and typically presents as solitary lesions.(Al-Ghadeer and Edward 2017) It is not prone to malignant transformation and has an excellent prognosis.(Tellechea *et al.* 2015) A case of perineural invasion has, however, been previously described.(Stern and Stout 1979) Treatment is usually geared toward cosmetic improvement.(Bharti *et al.* 2014) Simple surgical excision is curative in most cases and recurrence of the lesions at the primary post-excision is rare.("Trichofolliculoma: Background, Pathophysiology, Epidemiology" 2019; Lee *et al.* 2023)

We report a case of a rare presentation of multiple facial trichofolliculomas in an adolescent with childhood onset. Our aim is to add information to the limited data available on the presentation of trichofolliculoma.

Patient information

A 12-year-old female presented to our dermatology clinic at a tertiary institution in Ghana, with non-pruritic facial lesions of seven years duration .that had been present for seven years The lesions were occasionally painful and spared her trunk, limbs, and mucous membranes. They progressively increased in number and size over seven years (figure 1). There were no associated systemic symptoms. The patient had no chronic illness, and neither did any member of her family have similar lesions.

Clinical findings

Skin examination showed multiple skin-coloured nodules and papules of variable sizes (0.5 – 1.5 cm in diameter) with central pores confined to the face (figure 1). Eventhough the numerous lesions presented a challenge in individually counting them, we estimated the number of lesions to exceed 120. The hair, nails, and mucosae were intact. Examination of other systems was unremarkable. Before arriving at the histological diagnosis, we considered several potential differentials, including epidermal cyst, pilotrixoma, dermatofibromata, trichoepithelioma, and angiofibromata.

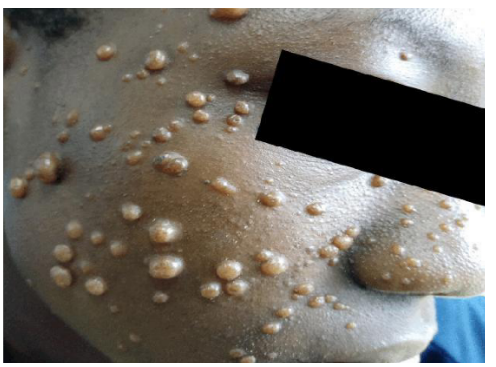

Figure 1. Multiple skin-coloured nodules and papules on the face.

Arrows point to skin-coloured nodules and papules of variable sizes (0.5 – 1.5 cm in diameter) with central pores confined to the face.

Timeline: Over a seven-year period, the lesions increased in number and size, with no spontaneous resolution or regression of any of the lesions.

Diagnostic assessment

An excision biopsy of a single lesion was done for histopathology. Since all the lesions appeared similar upon clinical examination, we chose to excise a single nodule for histological analysis primarily due to cosmetic considerations. Additionally, we opted for a well-defined lesion located away from major facial vessels.

Haematoxylin and eosin (H&E)-stained section revealed a polyp lined by an epidermis displaying basal melanin hyperpigmentation (figure 2). The core of the polyp showed a proliferation of dilated follicles which displayed follicular plugging. From the dilated follicles radiated numerous small follicles of variable maturity. These follicles showed a proliferation of basaloid cells with peripheral palisading. The surrounding stroma comprised a cellular, fibrous connective tissue stroma with no perifollicular stromal induction. These features were in keeping with the representation of trichofolliculoma.

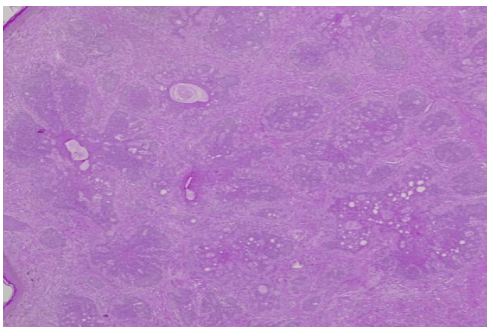

Figure 2. Histological features of Trichofolliculoma.

Haematoxylin and eosin (H&E)-stained section showing a polyp lined by an epidermis displaying basal melanin hyperpigmentation. The core of the polyp shows proliferation of dilated follicles which display follicular plugging. From the dilated follicles radiate numerous small follicles of variable maturity. These follicles show a proliferation of basaloid cells with peripheral palisading. The surrounding stroma comprised a cellular, fibrous connective tissue stroma with no perifollicular stromal induction.

Laboratory investigations including complete blood counts, renal function, and liver function tests were normal. Serological tests for HIV, HBsAg and HCV were non-reactive.

A diagnosis of trichofolliculoma was confirmed histologically.

Therapeutic intervention

We performed sequential therapeutic curettage of the lesions under local anaesthesia. Most of the lesions underwent curettage, with our selection based primarily on cosmetic concerns. Specifically, we targeted larger lesions that were deemed most cosmetically disruptive for the patient and located away from major facial vessels. Sequential therapeutic curettage was performed three times, once weekly over the course of one month.

Follow-up and Outcome.

After each procedure of curettage, healing was rapid (within four to five days) with no significant scarring (figure 3). No recurrence of the curetted lesions was noted during the six months of follow-up.

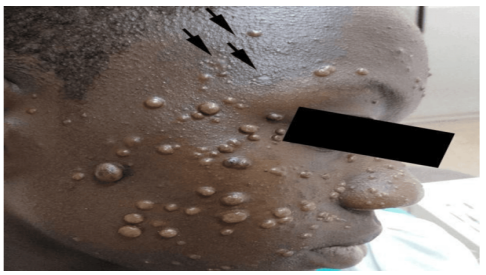

Figure 3. Post-curetage.

Arrows indicating the healed areas where curettage was performed

Informed Consent

Written informed consent was obtained from our patient and her legal guardian for the publication of this case report after ethical approval was granted by Kwame Nkrumah University of Science and Technology Committee on Human Research and Publication Ethics (CHRPE).

Patient's perspective

The patient was pleased with the results of the initial curettage and optimistic about the planned serial curettage.

DISCUSSION

Trichofolliculoma is a benign neoplasia of the hair follicles which usually presents in middle-aged persons (average age 45), with no precise gender or racial predilection. (El-Komy and Abdelkader 2020) Congenital trichofolliculoma is uncommon, with only one case reported.(Al-Ghadeer and Edward 2017) The onset of the current case however was in childhood, at age 5, which is unusual. The lesion tends to be located on the face, neck, and scalp.(Schulz and Hartschuh 1998) It rarely occurs in other areas of the body such as the genitalia, ear, and extremities, which was consistent with the locations of the lesions in this patient.(Choi, Lew, and Sim 2013) The occurrence of the lesions on the face of our patient is classical. Trichofolliculomas typically present as single lesions with few reports of multiple lesions.(Choi, Lew, and Sim 2013) Our patient presented with well-developed multiple facial trichofolliculomas, estimated to be approximately above 120 in number which is a rare presentation.

Although the exact etiology of trichofolliculoma is unknown,they are believed to represent abortive differentiation of pluripotent skin cells towards hair follicles. As seen in our patient, trichofolliculoma is not associated with systemic disease or other skin conditions and is hence classified as benign follicular tumours.(El-Komy and Abdelkader 2020). As seen in our patient, trichofolliculoma is not associated with systemic disease or other skin conditions and is hence classified as benign follicular tumours.

Trichofolliculomas encompass a group of neoplasms that share common histological and morphological features that resemble parts of the normal hair follicle. (Tellechea *et al.* 2015)

The differential diagnosis of benign follicular tumours includes trichofolliculoma, pilar sheath acanthoma, trichoadenoma, trichilemmoma, infundibuloma, proliferating trichilemmal cyst, dilated pore of Winer, trichoblastoma, pilomatricoma, trichodiscoma (fibrofolliculoma) and neurofollicular hamartoma. (El-Komy and Abdelkader 2020). For our patient, the potential differentials considered prior to histological diagnosis included epidermal cyst, pilotrixoma, dermatofibromata, trichoepithelioma, and angiofibromata.

Epidermal cysts were considered as a differential because they are equally discrete, freely moveable cyst often with a visible central puntum but these were absent in this patient. But they were less likely because, even though pilomatrixoma shares a common distribution and consistency as trichofolliculoma, stretching of the skin over the tumor with multiple facets and angles; the "tent sign" which is a pathognomonic sign for pilomatricoma was absent in our patient. Trichoepithelioma was also taken into consideration, although the lesions in this case lacked the keratinizing or horn cysts that are typical of the condition. Dermatofibroma, lesions are commonly hyperpigmented in blacks and pinching the lesion gently usually results in apparent downward movement of the tumour known as the dimple sign. This was not the case with the lesions in our

patient. Angiofibromas of tuberous sclerosis (TS) on the other hand, are usually reddish in colour and other signs of TS in the skin and other organs are present. This was not the case with our patient.

The characteristic histopathological features of trichofolliculoma include the presence of a central dilated cavity, lined with surface epithelium. The cavity is often continuous with the epidermis with several matured follicles radiating from the walls of the central cavity.(Ho and Bhawan 2017) There is also the presence of smaller infundibula cell lobules that irradiate from the main wall, helping to distinguish it from pilar sheath acanthoma.(Tellechea *et al.* 2015) The varied clinical and histopathological morphologies of trichofolliculomas are due to the evolutionary changes which occur with time. The three stages of trichofolliculomas include early, fully developed and late stages.(Misago *et al.* 2017)and that folliculosebaceous cystic hamartoma (FSCH

Trichofolliculomas can either present as sebaceous or folliculosebaceous cystic hamartoma depending on the evolutionary stage.(Ho and Bhawan 2017) Sebaceous trichofolliculoma is a variant with prominent sebaceous glands and sebaceous ducts within the lobules. Folliculosebaceous cystic hamartoma, on the other hand, is characterized by a more prominent mesenchymal component. It typically has no hair shaft formation or epidermal connection. (Wu 2008) The histological features of our patient's lesions seem to be consistent with folliculosebaceous cystics harmatoma since no hair shafts were seen and a more prominent fibrous stroma was noted.

Dermoscopy examination may be a useful diagnostic tool. The typical dermoscopic features are called "yellow clods". The presence of keratotic plugs within the follicular structures enlarges the follicular openings and gives rise to partially confluent white to yellow clods.

Although simple surgical excision is considered the standard of treatment (Lee *et al.* 2023), we opted to do curettage under local anaesthesia because of the number of lesions. Curettage was performed instead of simple excision because simple excision will require suturing. Considering the number of lesions and the facial distribution curettage offered better chances of rapid healing and cosmetic outcome.

CONCLUSION

Although rare, multiple trichofolliculomas can present in young patients. Histological diagnosis is key in the management, as it also helps to rule out similar conditions that may be malignant or prone to malignant transformation. Curettage under local anaesthesia could be explored as an alternative treatment modality in patients presenting with multiple lesions.

Conflict of interest

The authors declare no conflict of interest.

Authors contribution

All authors contributed to the patient's care and follow up as well as preparation of this manuscript.

REFERENCES

Al-Ghadeer, Huda, and Deepak P. Edward. 2017. "Congenital Sebaceous Trichofolliculoma of the Upper Eyelid." Ophthalmic Plastic and Reconstructive Surgery 33 (3S Suppl 1): S60–61. https://doi.org/10.1097/IOP.0000000000000698.

Bharti, Jyotsna Naresh, Biswajit Dey, Parul Gautam, Parth Desai, and Vinay Kamal.

2014. "Trichofolliculoma Presenting as Lobulated Mass: A Rare Presentation." International Journal of Trichology 6 (1): 19–20. https://doi.org/10.4103/0974-7753.136750.

Choi, Chang-Min, Bark-Lynn Lew, and Woo-Young Sim. 2013. "Multiple Trichofolliculomas on Unusual Sites: A Case Report and Review of the Literature." International Journal of Dermatology 52 (1): 87–89. https://doi.org/10.1111/j.1365-4632.2011.05120.x.

El-Komy, Mohamed Hm, and Heba A. Abdelkader. 2020. "Congenital Trichofolliculoma: A Very Rare Presentation." Dermatology Online Journal 26 (7): 13030/qt4g69d9zc.

Ho, Jonathan, and Jag Bhawan. 2017. "Folliculosebaceous Neoplasms: A Review of Clinical and Histological Features." The Journal of Dermatology 44 (3): 259–78. https://doi.org/10.1111/1346-8138.13773.

Lee, Ho Young, Eun Kyung Kim, Hyun Seung Choi, and Junhui Jeong. 2023. "Trichofolliculoma in the Auricle." Ear, Nose, & Throat Journal 102 (3): NP101–3. https://doi.org/10.1177/0145561321995599.

Misago, Noriyuki, Shin-Ichi Ansai, Takaya Fukumoto, Takashi Anan, Tetsunori Kimura, and Tomokazu Nakao. 2017. "Chronological Changes in Trichofolliculoma: Folliculosebaceous Cystic Hamartoma Is Not a Very-Late-Stage Trichofolliculoma." The Journal of Dermatology 44 (9): 1050–54. https://doi.org/10.1111/1346-8138.13842.

Pinkus, Hermann, and Richard L. Sutton. 1965. "Trichofolliculoma." Archives of Dermatology 91 (1): 46–49. https://doi.org/10.1001/archderm.1965.01600070052005.

Schulz, T., and W. Hartschuh. 1998. "The Trichofolliculoma Undergoes Changes Corresponding to the Regressing Normal Hair Follicle in Its Cycle." Journal of Cutaneous Pathology 25 (7): 341–53. https://doi.org/10.1111/j.1600-0560.1998.tb01757.x.

Stern, Jere B., and David A. Stout. 1979. "Trichofolliculoma Showing Perineural Invasion: Trichofolliculocarcinoma?" Archives of Dermatology 115 (8): 1003–4. https://doi.org/10.1001/archderm.1979.04010080061029.

Tellechea, Oscar, José Carlos Cardoso, José Pedro Reis, Leonor Ramos, Ana Rita Gameiro, Inês Coutinho, and António Poiares Baptista. 2015. "Benign Follicular Tumors." Anais Brasileiros de Dermatologia 90 (6): 780–98. https://doi.org/10.1590/abd1806-4841.20154114.

Trichofolliculoma: Background, Pathophysiology, Epidemiology." 2019, November. https://emedicine.medscape.com/article/1060303-overview.

Wu, Yu-Hung. 2008. "Folliculosebaceous Cystic Hamartoma or Trichofolliculoma? A Spectrum of Hamartomatous Changes Inducted by Perifollicular Stroma in the Follicular Epithelium." Journal of Cutaneous Pathology 35 (9): 843–48. https://doi.org/10.1111/j.1600-0560.2007.00914.x.