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Mini-Review

Syringocystadenoma Papilliferum at an Unusual Location: a Case Report

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Abstract:

A case of 15year old female presented with lesion over back since childhood, with occasional bleeding and oozing from lesion without any associated systemic complaints. There were multiple verrucous coalescing papules forming plaque with overlying erosion present over left lower back- diagnosed provisionally as angiokeratoma circumscriptum and was biopsied. Histopathology revealed findings consistent with Syringocystadenoma papilliferum. Surgical excision was done and closed with rotation flap. Syringocystadenoma is benign cutaneous adnexal tumor presenting clinically with many morphologies such as warty papules, nodules, plaques with oozing of serous material. Lesion is usually seen in head and neck area in most cases however can also occur on extremities, buttocks, anogenital region. It is characterized by multiple invaginations from skin surface in association with hair follicles lined by cuboidal to columnar epithelium on luminal aspect and myoepithelial cells on outside. There is papillary architecture and dermal ductal component.

Key words: Syringocystadenoma papilliferum; adnexal tumours; benign cutaneous tumours

Introduction

Skin appendageal tumours are rare tumours which either arise from or differentiate towards pilosebaceous apparatus, eccrine or apocrine morphology. These are more predominantly found in scalp and anogenital region given more density of pilosebaceous apparatus there. Pilar component predominates over scalp whereas glandular component tends to predominate over face. These present with vague clinical features and diagnosed only after histopathological characterization.

Case report

15year old female came with complaint of lesion over back since childhood. It was asymptomatic and pea sized to begin with, gradually increased in size over last 10-12 years. There is history of occasional bleeding and oozing from lesion. There are no systemic symptoms. On examination, patient had multiple skin-coloured papules coalescing to form plaque that was well circumscribed and had few eroded areas on lower left back. Few areas showed signs of spontaneous resorption. She was provisionally diagnosed as? angiokeratoma circumscriptum? lymphangioma circumscriptum. She was investigated further – here hemogram and blood biochemistry were unremarkable. Punch biopsy was taken and histopathological examination showed crateriform invaginations connected with surface epidermis that are lined by double layer of epithelium – luminal epithelium being apocrine and basal being cuboidal. On the floor of invagination, there were many papillary projections forming solid areas. Surrounding dermal tissue showed mixed infiltrate of lymphocytes, plasma cells and neutrophils. Thus histopathological diagnosis of Syringocystadenoma papilliferum was made and excision was planned in coordination with plastic surgery department. Lesion was excised and wound was closed with triangular rotation flap. The excised lesion measured 5X3X1 cm and on histopathology, confirmed diagnosis of Syringocystadenoma papilliferum without any cancerous changes.

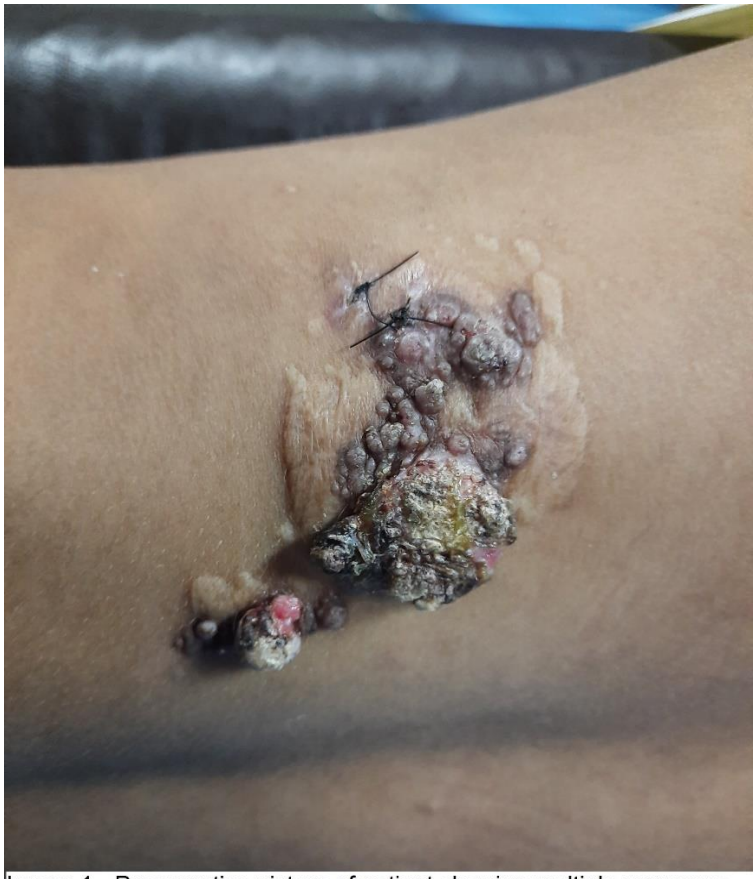

Image 1 - Preoperative picture of patient showing multiple verrucous papules coalescing to form plaque with erosion and crusting over surface. Areas showing spontaneous resolution also seen.

Figure 1: *Preoperative picture of patient showing multiple verrucous papules coalescing to form plaque with erosion and crusting over surface. Areas showing spontaneous resolution also seen*

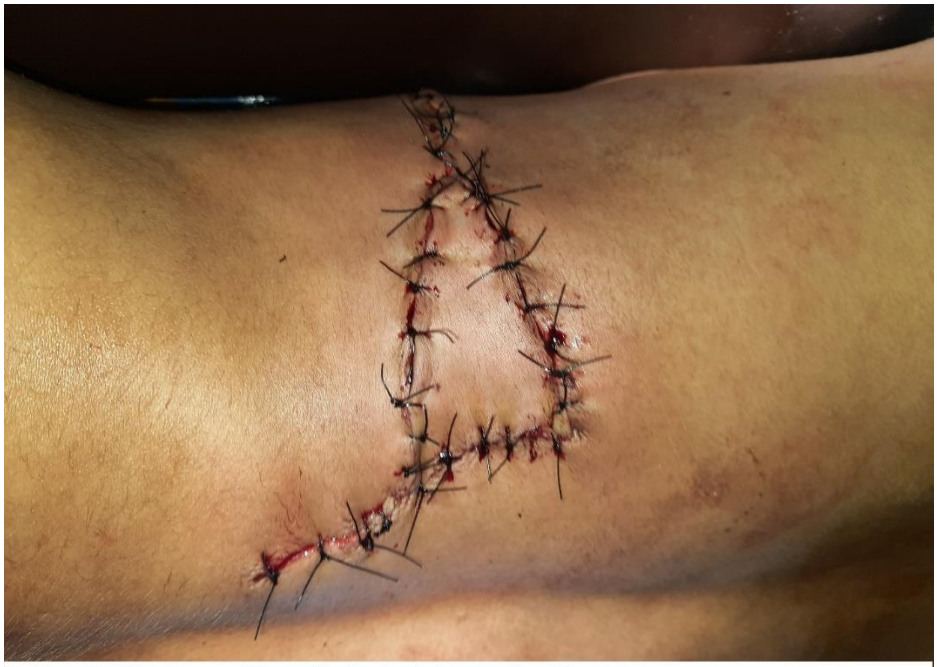

Image 2- Postoperative clinical picture of patient

Figure 2: *Postoperative clinical picture of patient*

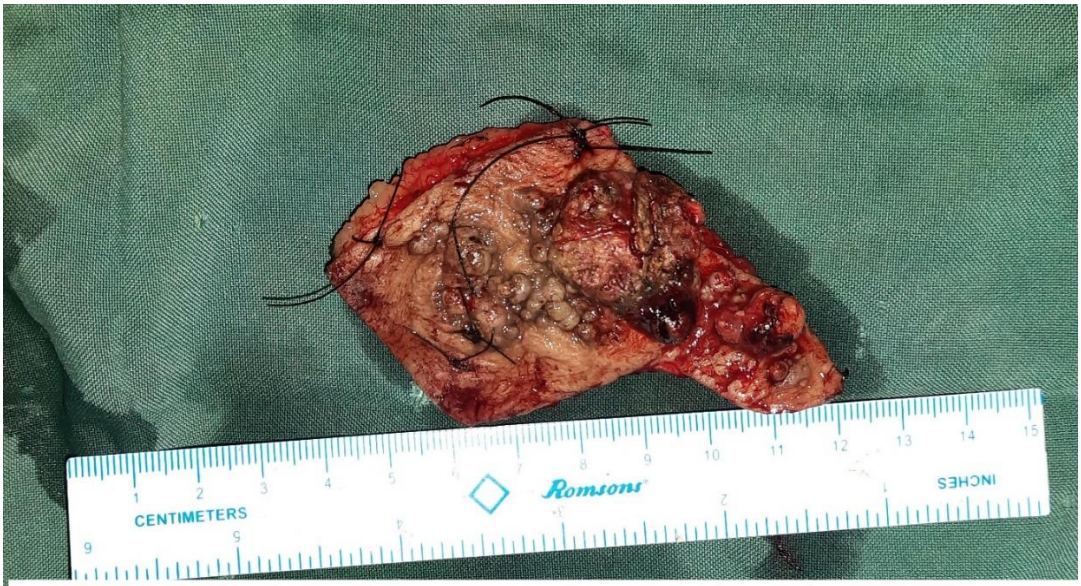

Image 3- excised specimen measuring aproximately 5X3X2 cm

Figure 3: *Excised specimen measuring approximately 5X3X2 cm*

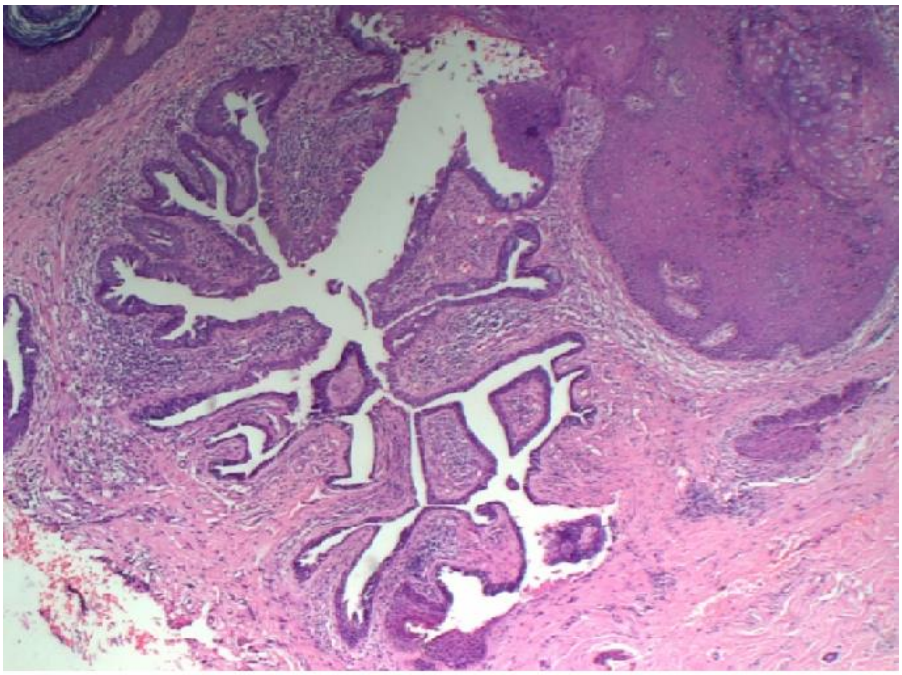

Image 4- Histopathology showing invagination from epidermis with apocrine morphology, with papillary projections toward base and few solid areas. There is inflammatory infiltrate composed of plasma cells, neutrophils surrounding the invagination.

Figure 4: *Histopathology showing invagination from epidermis with apocrine morphology with papillary projections toward base and few base and few solid areas. There is inflammatory infiltrate composed of plasma cells, neutrophils surrounding the investigation.*

Discussion

Syringocystadenoma papilliferum is a benign adnexal tumor that arises from glandular ductal epithelium. Its origin is uncertain- it's considered to arise from pluripotent appendageal cells or apoeccrine glands [1]. It can occur as an isolated tumor or as a secondary growth arising in Nevus sebaceous [2]. It occurs commonly in children and adolescents as acquired lesion, but can be congenital in rare cases [3]. In 75% cases it

occurs in head and neck region [4]. Other areas involved are groins, buttocks, extremities, anogenital areas [2]. In our case, the lesion was on left side of lower back.

Syringocystadenoma can present with many diverse morphologies – small grouped papules, nodules, warty outgrowth, crusted, oozing, moist fleshy nodules, plaques, papillomatous, cauliflower like or hyperkeratotic outgrowths [2]. Moist lesion can discharge fluid that can range from serous to thick serosanguinous and malodorous [5].

Histopathological features of early lesions branching glandular proliferations associated with infundibula of hair follicles- glandular component being composed of luminal cuboidal or columnar epithelium and peripheral myoepithelial cells. Well-formed papillae are rare in early lesion. Fully developed lesions will show a prominent papillary architecture with dermal ductal component [2].

Syringocystadenoma papilliferum can get transformed in syringocystadenocarcinoma – 4 such cases have been reported till date [6]. Basal cell carcinoma can develop in about 10% cases [7].

As with other benign adnexal tumours, simple excision is treatment of choice. However when lesions are multiple and associated with genetic syndromes, excision may not be feasible for all the lesions, in such cases, shave excision may be undertaken [8]. Other modalities of treatment are radiofrequency ablation, CO2 laser ablation.

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