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Syringocystadenoma papilliferum on the loin: An unusual case

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Abstract

Syringocystadenoma Papilliferum is a rare benign sweat gland tumour of varied presentation. We treated such a case in an eight-year female child presenting with an accelerated enlargement of a congenital polypoidal skin mass in the left loin with pruritus, ulcer, malodorous discharge, and cellulitis and left axillary lymphadenopathy. Our clinical diagnosis was Naevus Sebaceous transformed into Basal Cell Carcinoma with cellulitis of skin around. After subsidence of cellulitis with Flucloxacillin, and keeping it on, we performed excision biopsy of the mass and flap reconstruction. However, on the fourth postoperative day wound suppuration and wound dehiscence occurred that was cured with Ceftriaxone as per culture report and sensitivity results of the pus. As the wound became granulating, we suggested skin graft that the patient denied because of having an additional wound. As a result, there was a delay in wound healing and formation of a significant scar. Histopathology evaluation of lesion confirmed the diagnosis of Syringocystadenoma Papilliferum. From this study, we may conclude that loin is an unusual site of Syringocystadenoma Papilliferum. Such an ulcerated and rapidly enlarging lesion with lymphadenopathy clinically mimics a malignant transformation, but finally, histopathology evaluation of the mass confirms the diagnosis. This lesion with cellulitis of skin around has a potential risk for postoperative wound suppuration and delayed recovery in a one-step excision biopsy and flap reconstruction.

Keywords

Syringocystadenoma; loin; Ulceration; Cellulitis

Abbreviations

SCAP: Syringocystadenoma papilliferum; BCC: Basal cell carcinoma; SCC: Squamous cell carcinoma; NS: Naevus sebaceous; SCACP: Syringocystadenocarcinoma papilliferum; C/S: Culture and Sensitivity; FNAC: Fine needle aspiration cytology; SSI: Surgical Site Infection.

Introduction

Syringocystadenoma Papilliferum (SCAP) is a benign growth of the sweat gland of unknown aetiology and unexplained prevalence [1]. Fifty per cent of it is present at birth. It mostly appears de novo as a solitary nodule, nodular plaque, linear nodule, and polypoidal skin mass, and rarely in association with Basal Cell Carcinoma (BCC), Squamous Cell Carcinoma (SCC) and Naevus Sebaceous (NS) [2-4]. It has a potential risk of transforming into SCC, BCC and Syringocystadenocarcinoma Papilliferum (SCACP). So, an early excision biopsy is necessary [1-4]. It is usually asymptomatic though some patients complain of pain and pruritus [3,5]. An accelerated growth, pain, ulcer, pruritus and lymphadenopathy make suspicion of malignant transformation, and histopathology evaluation confirms the diagnosis [4,5]. It arises commonly in head and neck (70%), thigh, trunk, eyelid, cervix, Mons pubis and ear lobules are infrequent sites [2,4,5]. There is no example of SCAP on the loin in the literature. We presented here such a case with useful clinical information.

Case Report

A Bangladeshi mother attended us with her eight-year daughter having a congenital polypoidal skin lesion on left loin showing an accelerated enlargement, ulceration, discharge and erythema of skin around, pain, and difficulty in lying on the back. None of her family members suffered from a similar disease.

During the examination, her body weight was 20 kg, pulse 90 beats/m and body temperature 100^0F. There was a mildly tender polypoidal skin lesion on the left loin, measuring 5cm x 2cm x 2cm with malodorous discharge, and ulcer with overlying crusting and erythema of skin around (Figure 1a & 1b). Examination of axilla revealed a small tender, mobile lymph node to the left side. All other systems appeared normal. We suspected it for a naevus sebaceous transformed into Basal Cell Carcinoma.

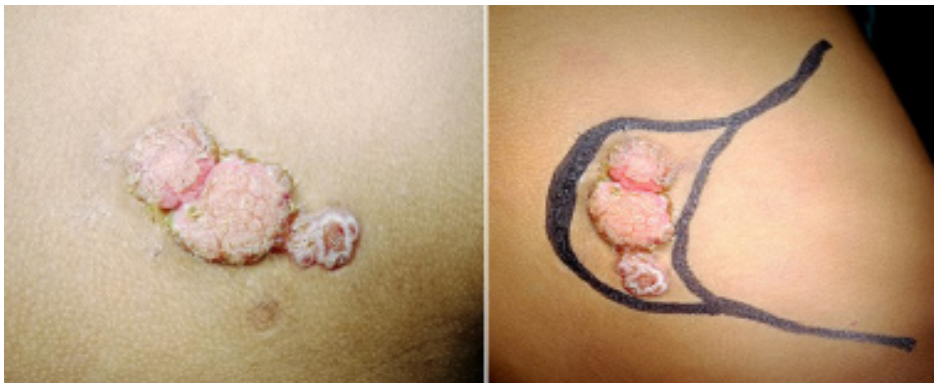

Figure 1: (a,b) The photographs demonstrating a polypoidal skin lesion in the loin with ulcers and crust and marking for excision and flap reconstruction.

The routine investigations were normal. We prescribed oral Flucloxacillin (125 mg four times a day) for seven days that cured cellulitis and fever. Keeping Flucloxacillin on, according to the demand of the patient and her parents for early removal of the mass, we excised it deep at the subcutaneous level and performed flap reconstruction. Histopathology evaluation showed cystic invagination extending into the dermis and composed of papillary projections lined by double layers of columnar epithelium with actual decapitation of secretion and dense lymphoplasmacytoid stromal infiltration (Figures 2a,2b,2c & 2d). The excision margin was free of tumour. The diagnosis was SCAP.

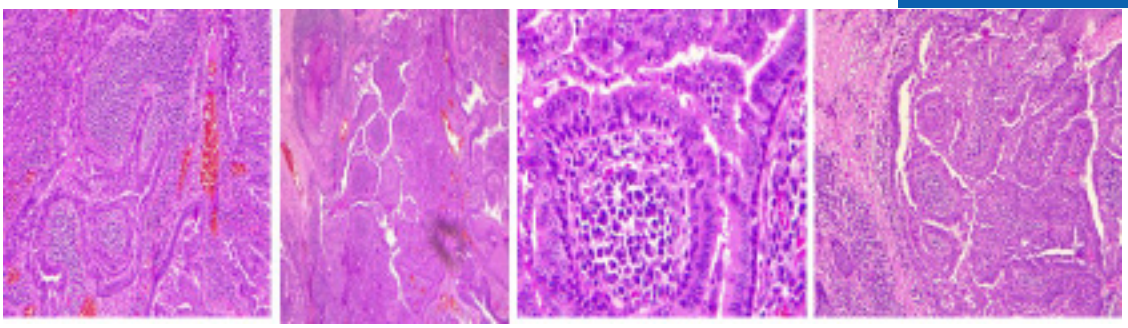

Figure 2: (a,b,c,d) Histological images of the tumour (Haematoxylin and eosin stain) showing cystic invagination extending into the dermis and composed of papillary projections and a dense lymphoplasmacytoid stromal infiltration.

On the fourth postoperative day, we detected wound suppuration and dehiscence. To know more about the bacterial growth, we performed culture and sensitivity (C/S) that showed Staphylococcus aureus highly sensitive to Ceftriaxone. With Ceftriaxone (50 mg/kg/day in a 12 hourly doses for seven days) suppuration and lymphadenopathy subsided, and the wound became granulating. We suggested skin graft, but the patient denied. As a result, there was a delay in healing and formation of a big scar (Figure 3a & 3b). Follow-up at six months showed no recurrence.

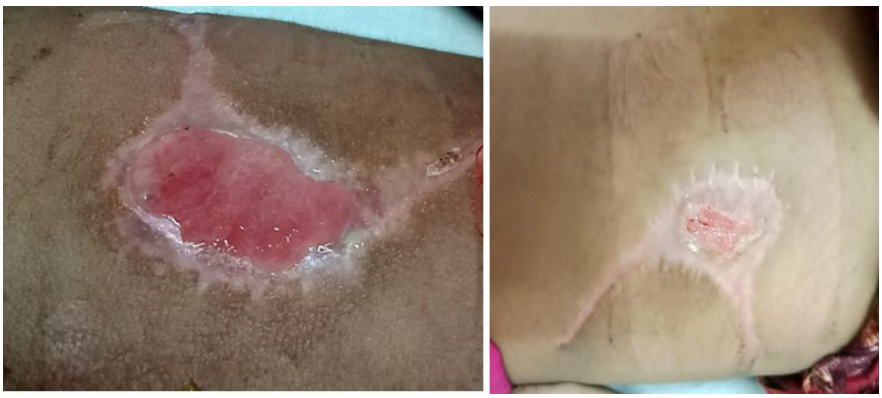

Figure 2: (a,b) Photographs showing healing wounds with evidence of flap reconstruction failure.

Discussion

Stokes first described SCAP in 1917. Review of literature showed that there are three hundred case reports until today [6]. Therefore, this is a unique example. The exact histogenesis of the disease is unclear. However, there is a suggestion of pluripotent stem cells [6]. Usually, it is asymptomatic, and perhaps, for this reason, the patient attends late for treatment. Diagnosis of clinical examination is difficult because of the varied presentation. Naevus Sebaceous, Hidradenoma Papilliferum, Papillary Eccrine Adenoma, and Warty Dyskeratosis are common differential diagnoses. Sometimes it presents with rapid enlargement, ulcers, discharge, pruritus and lymphadenopathy mimic a BCC or SCC. Histopathology evaluation is necessary to confirm the diagnosis and present case is a good example. As it has a potential risk of transforming to SCACP or SCC or BCC, early treatment is necessary. Comprehensive excision biopsy and flap reconstruction is the standard treatment [7-10]. Excision and flap reconstruction is usually a one-step procedure. However, there are examples of two-steps operation in suspicion of incomplete excision [11]. In this procedure, step-1 is the excision biopsy and step-2 is the flap reconstruction after confirmation of disease-free margin by histopathology evaluation [11]. In our case, we performed a one-step operation where postoperative wound infection occurred, and we think that it is related to preoperative infection of the lesion. In litera-

ture, there is no example of SCAP on the loin. Therefore, ours is the first case. Due to manoeuvre in the daily activities, the lesion on pressure areas such as loin poses a potential risk for sore and infection, causing pain, fever and lymphadenopathy, which mimics a BCC / SCC. Ultrasonography is a good tool to delineate lymphadenopathy. In fact, in this case, on clinical examination we detected enlarged tender lymph node to the left axilla, and we suggested Fine Needle Aspiration Cytology (FNAC) an excellent tool for diagnosis in this situation. However, for the unwillingness of the patient, we could not do it. As the patient asked for an early removal so as a preventive measure of SSI (Surgical Site Infection), we also could not do preoperative C/S of discharge and performed excision biopsy and flap reconstruction as soon as cellulitis subsided.

Conclusion

The presented case SCAP was on the left loin, an unusual site. Though it clinically mimicked a malignant lesion, in fact a benign one was confirmed by histopathology evaluation. Excision biopsy and skin flap reconstruction in one-step was complicated with postoperative wound suppuration, which might be related to preoperative infection of the lesion.

Learning Points

- Loin is an unusual site of Syringocystadenoma Papilliferum.
- Syringocystadenoma Papilliferum on pressure area, such as loin, has a potential risk for sore, cellulitis and SSI.

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References

1. Sangma MB, Dasiah SD, Bhat RV. Syringocystadenoma Papilliferum of the Scalp in an Adult Male – A Case Report. J Clin Diagn Res. 2013; 7: 742–743.

2. Goyal N, Aggarwal I, Bhargava S. Cytological diagnosis of syringocystadenoma papilliferum a rare adnexal tumour. J Cytol. 2015; 32: 130-131.

3. Dong Xu, Tianan Bi, Huanrong Lan, Wenjie Yu et al. Syringocystadenoma Papilliferum in the right lower abdomen: a case report and review of the literature. Onco Targets Ther. 2013; 6: 233-236. 10.2147/OTT.S4273PMID:

4. Barbarino S, McCormick SA, Lauer SA, Milman T. Syringocystadenoma Papilliferum of the Eyelid. Ophthal Plast Reconstr Surg. 2009; 25; 185-188.

5. Steshenko O, Chandrasekaran N, Lawton F. Syringocysadenoma papilliferum of the vulva: a rarity in gynaecology. BMJ. 2014: 5.

6. Google Analytics. Syringocystadenoma papilliferum – orphaned, https://www.orpha.net>consor>cgi-bin>oc_Exp

7. Shah PA, Singh VS, Bhalekar S, Sudhamani S, Paramjit E. Syringocystadenoma papilliferum: A rare case report with review of the literature. J Sci Soc. 2016; 43: 96-98.

8. Malhotra P, Singh A, Ramesh V. Syringocystadenoma papilliferum on the thigh: An unusual location. Indian J Dermatol Venereol Leprol. 2009; 75: 170-172.

9. Abak B, Gole P, Dongre A, Kharkar V, Jain S. Syringocystadenoma papilliferum of the cervix presenting as vulvar growth in an adolescent girl. Clinical and Experimental Dermatology, 2019; 44: 58–61.

10. Tseng MC, Amin B, Barmettler A. Eyelid Syringocystadenoma papilliferum : A novel presentation with a major review. Orbit. 2018, 6; 37(3): 171-174.

11. Brent AJ, Mota PM, Nebojsa A et al. Squamous cell carcinoma arising from syringocystadenoma papilliferum of the eyelid. Canadian Journal of Ophthalmology, December 2017, volume 52; issue 6: e235-e237.

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