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Solitary and Pedunculated Nevus Lipomatosis Cutaneous Superficialis of the Thigh– A Rare Case Report and Review of Literature

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Author's contribution

The sole author designed, analysed, interpreted and prepared the manuscript.

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Case Report

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ABSTRACT

Background: Nevus lipomatosus cutaneous superficialis (NLCS) is a rare benign hamartomatous idiopathic condition. The characteristic feature of this condition is the presence of mature adipocytes in the dermis. Two forms of NLCS are identified clinically vis-à-vis classical form and solitary form. The latter is seen in adults and is a rarer variant than the former.
Case Representation: This case report describes a solitary and pedunculated form of NLCS seen in the thigh of a young adult, a rare variant of NLCS. The swelling was excised surgically followed by good recovery with no recurrence.
Conclusion: Early recognition of this benign condition will result in less morbidity amongst the patients with regards to cosmesis especially when the lesion can recur.

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Keywords: Pedunculated nevus lipomatosus cutaneous superficialis; solitary variant; NLCS; lipofibroma.

1. INTRODUCTION

Nevus lipomatosus cutaneous superficialis (NLCS) is a rare benign hamartomatous idiopathic condition as described by Hoffman and Zuhrelle in 1921 for the first time [1]. Ectopic mature adipose tissue within the dermis is the key salient feature of this condition. Two clinical variants of NLCS are illustrated in the literature vis-a vis the classical form and solitary form. The former variant consists of multiple pedunculated cerebriform shaped nodules, that resembles a plaque most often. The second clinical variant of NLCS is relatively a rare form, presenting as a solitary sessile papule or nodule [2]. Very few cases on solitary form of NLCS have been published in the literature. A solitary, pedunculated neoplasm can be easily misdiagnosed as papilloma or skin tag. This case report describes a solitary and pedunculated NLCS in a young adult, a rare variant of NLCS with review of the literature.

2. CASE REPORT

A young female aged 27 years presented to our department with a four-year-old history of a swelling on the inner side of left upper thigh. The swelling had originally appeared without any trigger and gradually increased to the present size over the four years. There was no significant family history. Physical examination showed a pedunculated, solitary nodule measuring 40×40 mm. The prominent stalk was 10 mm long. The swelling was skin-colored, nodular in appearance, soft in consistency, non-tender, with no sinuses or ulceration (Fig. 1A). Surrounding skin appeared normal. No other associated swellings were noted. Fine needle aspiration of the swelling reported scattered mature adipocytes with many fibrous strands against a hemorrhagic background. Under local anesthesia, an elliptical incision was marked around the stalk and deepened to completely excise the swelling (Fig. 1B). Incision was closed using non-absorbable sutures (polyethelene 4-0). Histopathological examination (HPE) of the swelling showed infiltration of the epidermis and dermis with chronic inflammatory cell infiltrate. The dermis showed presence of mature adipocytes were seen in lobules separated by fibrocollagenous stroma. Blood vessels were also seen in the stroma. No dysplastic changes present (Fig. 2). The findings were suggestive of NLCS, solitary pedunculated variant. Post-operative period was uneventful. Sutures were removed in two weeks after the surgery. One year follow-up showed no recurrence.

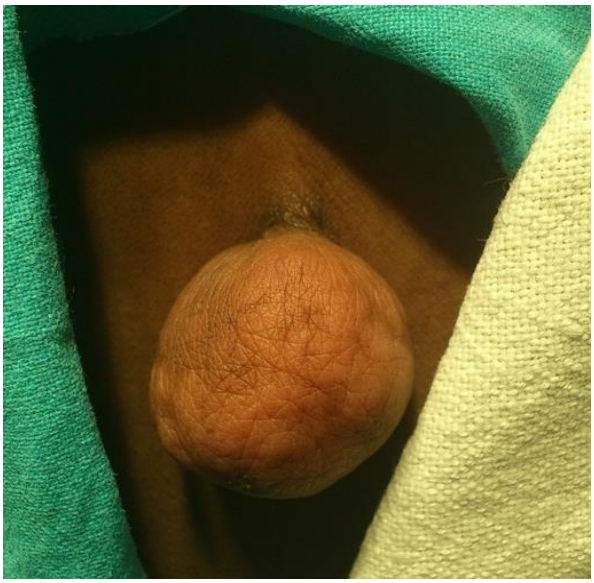

Fig. 1A. Pre-operative image of the pedunculated, solitary nodule measuring 40×40 mm with a prominent stalk measuring 10 mm long. The swelling was skin-colored, nodular in appearance, with no sinuses or ulceration

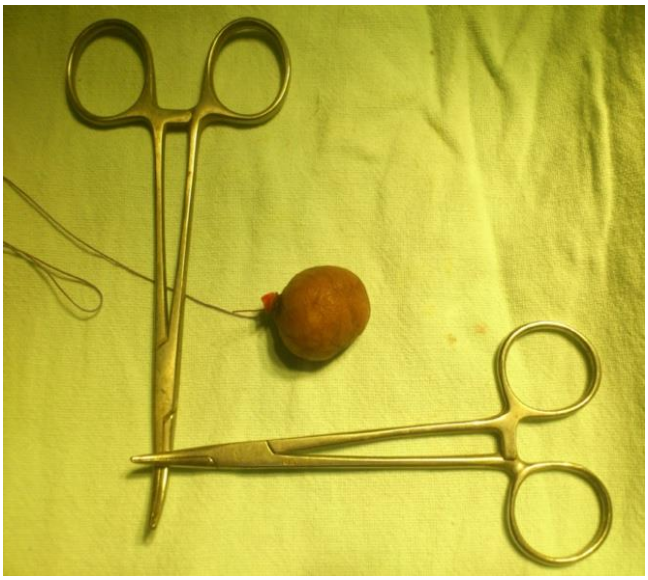

Fig. 1B. Excised swelling with the stalk

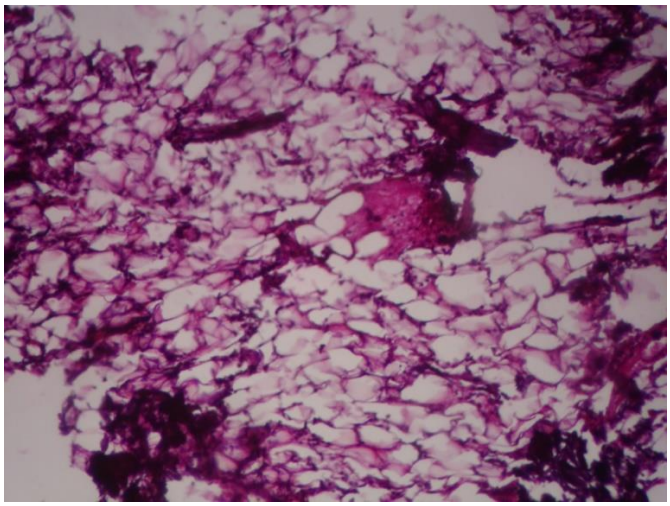

Fig. 2. Low power view showing hyperkeratosis, irregularly organized connective tissue and presence of mature adipocytes in the dermis

3. DISCUSSION

The solitary form of nevus lipomatosus cutaneous superficialis is described as “pedunculated lipofibroma” in the literature which was suggested by Mehregan et al [3]. It is a rare, benign connective tissue neoplasm. Ectopic adipose tissue in the dermis is the characteristic feature of this slow growing, solitary variant of NLCS. These histological features are similar to the classic variant of NLCS [1]. The lesions in the classic type of NLCS are seen since birth or develop later, during the first three decades of life. The common sites of occurrence are usually the gluteal region, lower back, and the thighs. The solitary variant can be either nodular, papular or pedunculated which usually occurs later in life (over 20 years of age) and is found in unusual sites like axilla, knee, eyelid, nose, ear/pinna, clitoris, scrotum, or skin of the scalp [4-6].

NLCS are slow growing and asymptomatic in nature. However, large lesions tend to ulcerate with superimposing bacterial infections [2]. A large NLCS has caused compressive ulnar neuropathy, as reported by Tunce et al [7]. There is no documented genetic predilection, but one

study has reported the association of 2p24 deletion [8].

The pathogenesis of NLCS is unknown. Various theories have been mentioned in the literature. One of the theory is that the subcutaneous adipose tissue is displaced into the dermis due to the degenerative changes noted in the collagen and elastic tissues. Another theory believed is that lipoblasts are noted in the dermis which differentiates to adipose cells from the walls of dermal capillary vessels [9].

The main histological abnormality in either type of NLCS is ectopic fatty tissue in the upper dermis, which can be distinctively differentiated from the normal subcutaneous fat. The connective tissue are organized irregularly with dermal blood vessels surrounded by adipocytes [10,11]. Nogita et al, in their large case series of 32 cases demonstrated increased deposition of mucopolysaccharides in the reticular dermis and fatty tissue after staining with alcian blue [12].

Clinically and histologically, the differential diagnosis for a pedunculated NLCS includes other benign papillomas, including acrochordons, seborrhoeic keratosis, nevocellular nevi, verrucae, neurofibromas, fibroepithelioma of Pinkus, eccrine poroma, focal dermal hyperplasia (Goltz syndrome) [4]. HPE will form the key contributor to the final diagnosis.

Complete excision of the lesion forms the mainstay treatment of pedunculated NLCS, for both therapeutic and cosmetic reasons. Excision of large sessile plaques may result in soft tissue defects. These defects will necessitate either skin grafts or flap surgery. The latter may be combined with tissue expansion. The non-surgical options include cryotherapy, carbon dioxide ablative laser, corticosteroids topical applications, intralesional phosphotidylcholine and sodium deoxycholate injections [13-15]. Recurrences have been noted following these modalities when surgical excision becomes an absolute necessity [14]. Although there are no reports in the literature about any malignant transformation of pedunculated lipofibroma, it is prudent to examine these swellings for malignancy considering the incidence being rare.

4. CONCLUSION

Nevus lipomatosus cutaneous superficialis is a rare skin pathology and solitary, pedunculated form appears to be a rarer variant. Early recognition of this benign condition will result in less morbidity amongst the patients with regards to cosmesis. Large, pedunculated lesions are susceptible to torsion, ulceration. The comprehensive review of the literature has suggested that surgical excision remains to be the gold standard treatment to prevent recurrence and early intervention will prevent the need for large reconstructive options unless indicated.

CONSENT

Informed consent was obtained from the patient for the use of photographs for publication. The consent has been formally documented in the medical record.

ETHICAL APPROVAL

As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Author has declared no known competing financial interests or non-financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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