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Case Report

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Nevus Lipomatosus Cutaneous Superficialia and its Association with Metabolic Syndrome

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ABSTRACT

Introduction: Nevus Lipomatosus Cutaneous Superficialis (NLCS), is the name given by Hoffmann and Zurhelle to a nevoid anomaly consisting of grouped papules and nodules which on histologic examination showed the presence of ectopic fatty tissue in the corium. It is considered as a rare entity with unknown etiology.

Methods: This case series includes clinical data and histopathological findings of nine cases of histopathologically documented NLCS who presented to the district General Hospital Kilinochchi over a period of 2 years (January 2021 to December 2022).

Discussion and Conclusion: NLCS is a rare and benign condition, which should be differentiated from other skin conditions as it commonly mimics several other benign skin conditions. 55.5% (5/9) were females and the duration of the lesions was ranging from 3 to 25 years. The size was seen to vary from 03 x 02 cm to 10 x 07 cm, located at the buttocks (4/9), shoulder (2/9), back of the neck (1/9), back of the chest (1/9), thigh (1/9). The mean age of the patients was 32.2 years ranging from 14 to 45 years. The majority of patients show high FBS (57.1%) and total cholesterol levels (100%). No familial tendency was reported. There is a significant association between this condition with FBS and with total cholesterol raising the possibility of an association of NLCS with metabolic syndrome.

Keywords: Nevus Lipomatosus Cutaneous Superficialis, Pedunculated, Fibrofolliculoma

Introduction

Nevus Lipomatosus Cutaneous Superficialis (NLCS), is the name given by Hoffmann and Zurhelle to a nevoid anomaly consisting of grouped papules and nodules which on histologic examination showed the presence of ectopic fatty tissue in the corium. It is considered as a rare entity with unknown etiology [1,2].

According to our knowledge, no published Sri Lankan case reports or case series have been found on this topic and according to the available published studies in the international publications, NLCS has been reported as a rarer entity. Only a few case reports and studies are available for reference. Most of these have been identified during the routine examination for histology in specimens obtained after excision surgery for various other reasons including cosmetic demand by patients.

Although this is described as a rarer entity in clinical practice, nine cases have been reported at our hospital over a period of 2 years, highlighting the need for awareness of this rare clinical condition with opening an assessment. Here, we have been trying to evaluate some clinicopathological features of this rare entity reporting a series of 9 cases of NLCS in a short span of twenty-four months.

Case Series

Retrospectively we collected clinical data and histopathological findings of nine cases of histopathologically documented NLCS who presented to the district General Hospital Kilinochchi over a period of 2 years (January 2021 to December 2022).

The demographic and clinical data were recorded from the accompanying Histopathology request forms. Slides and tissue blocks were retrieved. Haematoxylin and Eosin (H&E) slides were reviewed in all cases.

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The following clinical characteristics were recorded for analysis: age, gender, clinical features, histopathology, FBS and total cholesterol levels. The clinical profile of all the patients is given in Table 1.

Table 1: Clinicopathological Details of All Nine Patients

	1	2	3	4	5	6	7	8	9
Age at presentation	26	33	38	14	37	37	45	39	21
Sex	F	M	M	F	M	F	M	F	F
Duration	8	10	10	3	18	20	25	15	6
Site	L/ Buttocks	L/ Shoulder	R/ Thigh	Back of the neck	L/ Buttocks	R/ Buttock	Upper back of the chest	R/ Shoulder	L/ Buttocks
Size	6 x 12cm	4 cm	6 x 3 cm	2 x 8 cm	3 x 5 cm	10 x7 cm	5 x 3 cm	4 x 8 cm	1 x 3 cm
Pedunculated/Sessile	P	P	P	S	P	P	P	S	P
Multiple/ Solitary	M	S	M	M	M	S	S	M	M
FBS	95	-	142	-	128	134	126	108	96
Total Cholesterol	-	-	232	-	-	-	288	-	-

Figure 1: A single pedunculated mass with cribriform outer surface and yellow glistening cut-surface

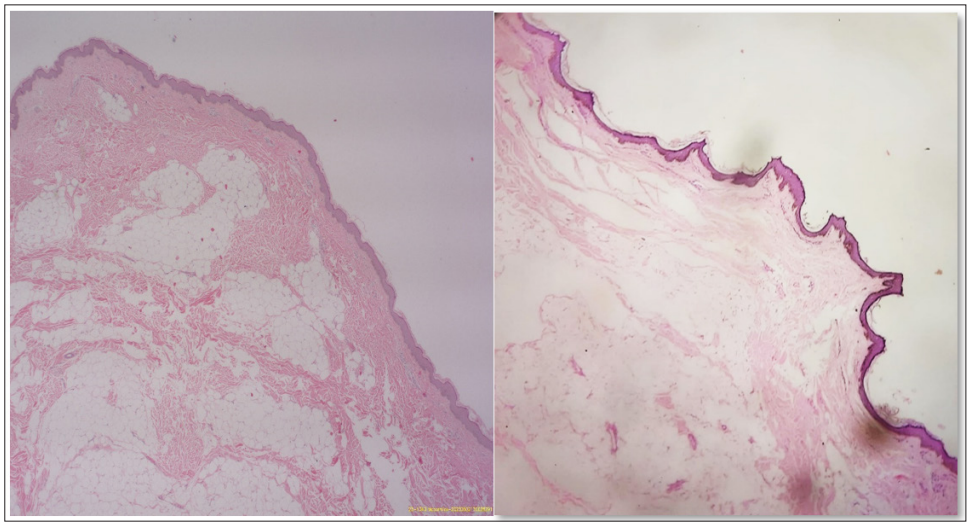

Figure 2: Attenuated epidermis with lobules of mature adipose tissue in the upper reticular dermis. (H & E x200)

Our nine patients had been distributed the ages ranged from 14 to 45. Out of nine patients, five were women and four were men. The duration of the lesions was ranging from 3 to 25 years. The shortest duration of the lesion was 3 years, but that patient was the youngest of the group.

The size was seen to vary from 03 x 02 cm to 10 x 07 cm, located at the buttocks (4/9), shoulder (2/9), back of the neck (1/9), back of the chest (1/9), Shoulder (1/9). The majority of lesions were multiple (6/9). Except for two cases, all the others were pedunculated lesions. Fasting blood sugar (FBS) levels were found in seven patients and patients 3, 5, 6 and 7 had diabetics. We were able to find the total cholesterol levels of only two patients: patients 3 and 7 and both showed high cholesterol levels than the upper normal limit. None of the patients reported a familial tendency for the same.

Discussion

NLCS is an uncommon benign cutaneous hamartomatous lesion. In 1921, NLCS was first described by Hoffmann and Zurhelle, in a 25-year-old man who presented with multiple soft nodules on the gluteal region. Clinically, two forms have been described: The classical (multiple) Hoffman-Zurhelle form and the pedunculated solitary form. Classical NLCS is usually present at birth or during the first two decades of life whereas, the solitary form of NLCS usually manifests in the third to sixth decades of life. However, the latter may be present at birth also. Though the pelvic girdle is the most common site for the classical form, the solitary form can occur at any site. Only a single study has reported a predominance of solitary form comprising 13 cases spanning 12 years [1-9].

These lesions are slow growing over several years and usually leave untreated as the patients do not have any symptoms other than cosmetic. Our study showed that patient 7 was presented after 25 years but the size was 5x3cm, while the largest mass obtained was from patient 6 and it had been identified 20 years ago. Further, the smallest lesion was seen in patient 4 and the duration was also the shortest among the group and that patient was the youngest. So, there may be a relationship between the size and the duration of the lesion. To date, Malignant transformations have not been reported and recurrences are also extremely rare.

Further, there is a clue that there may be a relationship between NLCS and metabolic syndrome as the majority of patients showed high FBS (57.1%) and total cholesterol levels (100%). A study done by Nogita et al noticed that 21% of their sample had diabetes mellitus. However, NLCS is an idiopathic anomaly. Family history, ethnicity or predominant sex involvement have not been described. Similarly, the role of chromosomal abnormalities in this condition is inconclusive. Though, a case report of a NLCS with a 2p24 deletion has been reported [9,2,10].

The histology of both clinical subtypes of NLCS shows characteristic ectopic mature adipose tissue in the reticular dermis [2,6] and usually extends to the papillary dermis. These fat lobules may or may not show continuity with subcutis. Other than the presence of fat cells, the dermis is normal but, in some conditions, increased amounts of dermal collagen and fibroblasts are seen. The number of blood vessels is also commonly increased and fat cells are also seen around them [11,4]. Epidermis is usually normal, but attenuation or hyperplasia can be seen. Even though the number of adnexal structures is reduced in NLCS, there are reported cases of co-existing pilar anomalies in the form of hypertrophic pilosebaceous units, hyper follicular fibrosis, fibrofolliculomas and trichofolliculomas [3].

The pathogenesis of NLCS is not yet determined. Some authors have suggested that it originates from adipocyte precursor cells located around blood vessels. However, in a study with electron microscopy, only mature lipocytes were observed inside the perivascular mononuclear infiltrates, and no lipoblasts or the transition from mesenchymal cells to lipocytes were observed. Another study reported the deletion of 2p24 in NLCS, indicating a role of genetic factors in development of NLCS [3,6,12]. However, the etiology remains unknown.

Because of the rare nature of this disorder, it is often misdiagnosed. It clinically mimics other benign skin conditions, including neurofibroma, nevus sebaceous, angiolipoma, trichoepithelioma and cylindroma. Histopathology is essential for the diagnosis. The need for awareness among healthcare providers of its clinical and histopathological features prompted us to share our experience with NLCS [2].

The treatment of choice is surgical resection, which is for cosmetic purposes only, given the benignity of the lesion treatment is not medically necessary. An early diagnosis may permit a more conservative resection of the lesion and less invasive reconstruction of the defect since the lesions can grow extremely large causing aesthetic concerns to the patient [12].

Conclusion

NLCS is a rare and benign condition, which should be differentiated from other skin conditions as it is commonly misdiagnosed. Therefore, physicians and surgeons should be aware of its clinical and histopathological features. Since the pathogenesis including environmental factors and associated genetic abnormalities of NLCS is not yet determined and as this rare skin condition has close relationship between metabolic syndrome this would open a flat form for research studies

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