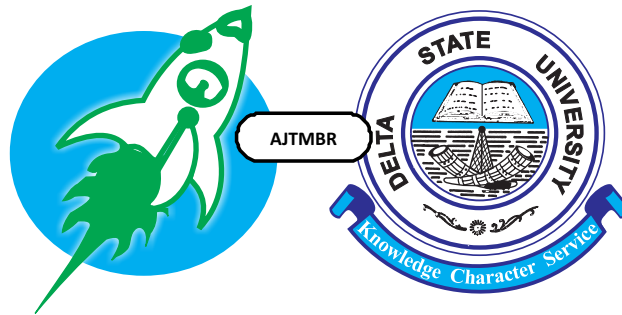


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Case Report

Lichen Sclerosus in Extremes of Age: A Report of Two Cases in Skin of Colour in a Secondary Health Facility in South-South, Nigeria

Sokunbi AE¹, Omenai SA²

ABSTRACT

Lichen sclerosus (LS) is a chronic dermatologic condition with a bimodal peak age of occurrence; it is encountered in pre-pubertal paediatric ages and post-menopausal populations. This dermatosis is thought to have an autoimmune basis and has been rarely reported in our environment. Although more common in adults, it can affect individuals of all ages. This case series presents two unique cases of Lichen sclerosus in a 4-year-old female and a 70-year-old female reported in persons with skin of colour, highlighting the variations in presentation, diagnosis, and management in these distinct age groups. This case series also aims to increase awareness about this condition which may likely have been misdiagnosed as other similar dermatoses; thus limiting the case detection rate of this dermatosis in African population.

Keywords: Lichen Sclerosus, Paediatric Age, Elderly, Skin of Colour, Nigeria.

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INTRODUCTION

Lichen sclerosus otherwise known as lichen sclerosus atrophicus is a relatively uncommon chronic inflammatory dermatosis that is often recurrent and fraught with the possibility of malignant association.¹⁻⁷ This dermatosis bears a female preponderance and has been described all over the world; especially in Caucasians.^{8, 9} However, there seems to be a dearth of reports of cases among blacks especially in this environment.^{3,8,9}

Lichen sclerosus bears several dermatologic manifestations and patterns with overwhelming occurrence in the genitals especially in post-menopausal women¹⁻⁸ with topical steroids being the gold standard for treatment.^{2,4}

CASE REPORT

CASE 1: A 4-year-old female who was noticed to have developed whitish atrophic lesions on the vulva with associated mild discomfort and itching. The lesion was noticed to have progressively increased in size over 3 months; necessitating the visit to the dermatologist. Further history revealed the appearance of a similar lesion on the trunk about a year prior, which resolved without treatment. There was no family history of similar lesions and no other history suggestive of autoimmune disease in this patient.

Examination revealed well-defined depigmented patches on the vulva spreading to the labia majora and perineum posteriorly; with a shiny, atrophic surface. The diagnosis of Lichen sclerosus was

confirmed by histology to exclude the possibility of other differential diagnosis.

Patient was commenced on emollients and

topical 0.05% clobetasol propionate with complete resolution of symptoms (Fig. 2) while the steroid was tailed off after one month.



FIG:1

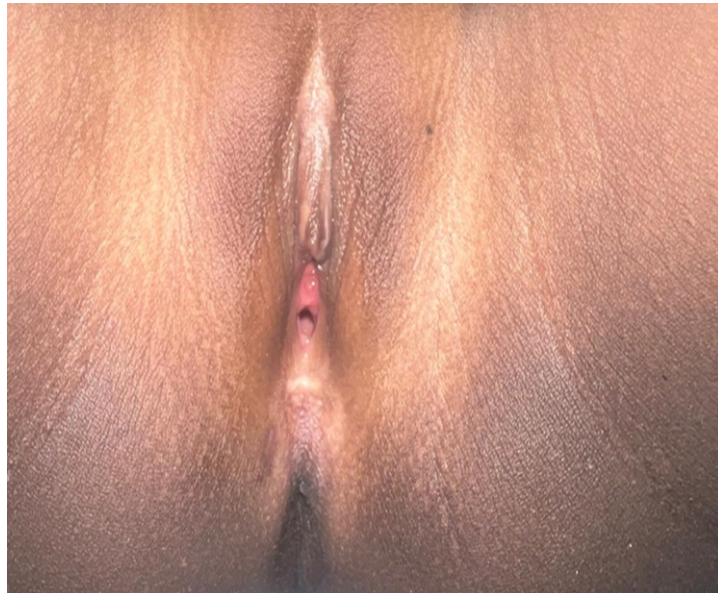


FIG:1

CASE 2: A 70-year-old post-menopausal female who noticed the appearance of white patches on the vulva area which grew larger; extending to involve the labia minora. There was associated marked discomfort, pruritus and pain on urination. Itching was severe with occasional associated soreness of the area. There was no history suggestive of weight loss or malignancy; though dyspareunia could not be ascertained as she had not been sexually active for many years.

Examination revealed a depigmented patch on the labia majora extending to the labia minora with associated background erythema.

Urinary tract infection was excluded after investigations and the diagnosis was confirmed by histology which also excluded possibility of malignant association. She improved remarkably after the commencement of topical 0.05% clobetasol propionate, emollients and oestrogen.

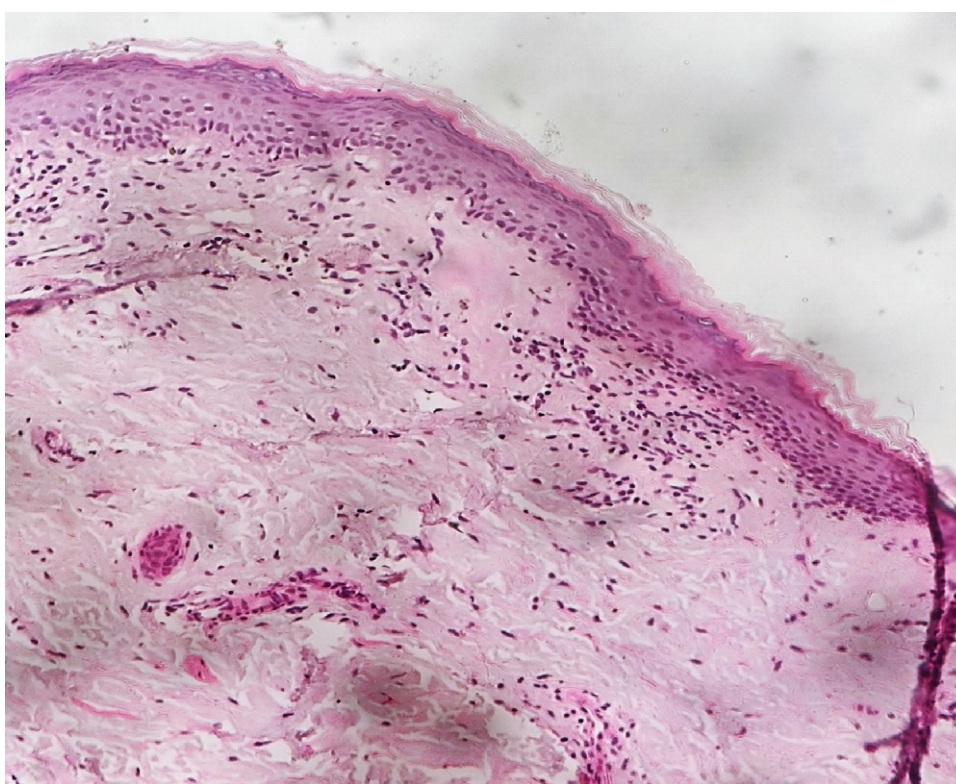


Fig 3: Sections show hyperkeratosis, epidermal thinning with loss of rete ridges with the interface dermal sclerosis intervening between mild to moderate lymphocytic inflammation. (H&E x200)

DISCUSSION

This review aims to evaluate previously published reports on the disease, focusing on clinical correlates such as clinical presentation, diagnosis, management, and outcomes. The results from these case reports varied in terms of the type of patients included in the study

(demographics), clinical presentation, management strategies, and outcomes.

Generally, regarding the demographics of the study, the majority of the case reports included females, with a high incidence among postmenopausal women. The ages of the patients

included in the study ranged from 15 years to 84 years.^{4,10} The affected areas noted in literature included extragenital areas (oral mucosa, hand, leg, breast, trunk, back, shoulder)^{1-4,10} and most commonly the ano-genital region¹⁻¹⁰

The disease can be asymptomatic as seen in most of the reports;^{4, 11} however, the most common presenting symptoms in the studies included itching, bleeding, white atrophic patches/plaques in the affected areas.^{4,7} Additionally, the common method of diagnosis in the studies included histopathological examinations and biopsy of the affected area.⁴

Lichen sclerosis being commoner in females bears a male to female ratio of 1:10 to 1:6 in different environments.¹³ In addition, it is commoner among adults, with fewer incidences reported in children. Its estimated incidence is 0.04–0.06% in children compared to 0.1–0.6% in adults.¹²⁻¹⁴ In postmenopausal women, the estimated incidence is 1:1000.²

Lichen sclerosis has been described in many ethnicities as a chronic inflammatory dermatosis of unknown aetiology¹⁻⁵ which is 10 times commoner in females^{8,10} with associated bimodal peak of occurrence.⁴ The condition has been described in about 3% of post-menopausal women,⁵ while in paediatric populations where it is thought to be under-diagnosed;² the reported prevalence is about 6- 10 %.^{5,6} Although few studies have reported Lichen sclerosis in skin of color, not many are from this environment. There appears to be an underreporting in our environment because some studies have reported contrary prevalence among black Americans.¹⁵ This study conducted among subjects with skin of colour, identified vitiligo-like Lichen sclerosis to be commoner in this population. This is similar with case 1 presented as the lesions were mostly perineal depigmented

patched that could be confused with Vitiligo, necessitating confirmation by histology. There is therefore a possibility that depigmented patches may be clinically diagnosed as vitiligo which coincidentally also responds to topical steroids. Also, the cases identified in this series corresponds with the classical epidemiological characteristics of Lichen sclerosis where the first case was a young pre-pubertal female and the other a post-menopausal woman.

The etiology of Lichen sclerosis is largely unknown. However, it has been associated with many factors such as hormones, trauma (Koebner phenomenon), infections such as; human papilloma virus, hepatitis C virus, *Borrelia* and asymptomatic mycobacterial infection, drugs, auto-immune conditions including thyroid diseases, Pernicious anaemia, vitiligo, psoriasis; and atopic dermatitis, and diabetes mellitus with the disease.^{2,4,8} Pre-pubertal Lichen sclerosis has been associated with auto-immune diseases in about 6.6% of cases.²

About 56% of cases reported in literature are known to carry familial association. A case report by Doulaveri et al. in 2013 suggested genetic role in the etiology of the disease; reporting vulvar Lichen Sclerosis in Monozygotic Twin Women. The affected areas were predominantly in the anogenital region, and there was no preceding trauma in the genital region.¹⁶ The most significant discovery in the study indicated that their father and mother suffered from various forms of Lichen sclerosis, such as balanitis xerotica obliterans and lichen sclerosis et atrophicus, respectively.¹⁶ Similarly, another study on 1052 patients in 2010 found that 12% of the participants had family history of LS.¹⁷ Additionally, the report was in line with two studies conducted in 1994 and 2006 also had cases of familial LS. Furthermore, a clear evidence of associated HLA markers in Lichen sclerosis was

reported in the literature.^{18, 19} The HLA- types include HLA –DQ 7 (seen in 60% of females), HLA- D7 (in 60% of females), 8 and 9; HLA-DR-12. Affected subjects were also found to test negative for HLA-DR17.^{2,4}

Other evaluated case reports involved the incidence of LS in extragenital areas.²⁰⁻²³ As discussed previously in this section, the disease was asymptomatic in these reports, and dermoscopy was useful in diagnosing the condition in a reported case where the lesion was present on the hand, while biopsy was used in the rest of the studies. After applying various treatment procedures, significant improvements were seen in the occurrence of Lichen sclerosus in the affected areas.⁴

Several clinical manifestations of the disease have been described in literature. The classical presentation is depigmented/ ivory-coloured lesion with associated skin atrophy which can be found in both genders.¹⁻¹⁰ The lesions of Lichen sclerosus are most commonly located on the anogenital region^{4,5} (even extending into the vagina in females) as seen in both cases reported. These lesions are often associated with itching, pain, discomfort, dyspareunia and itching in the vulva. 79% of post –menopausal women with lichen sclerosus have been reported to be saddled with chronic vulva pain.⁴ However, several reports of other patterns of the dermatosis have been described in other parts of the body such as the face, neck and trunk; with generalized lesions also described in literature.^{3,4, 10} Asymptomatic plaques on the breasts in both males and females have also been noted.¹¹ Rarer forms of the dermatosis have been reported in the form of bullous, haemorrhagic lesions.^{10,23}

Asides females, the dermatosis has been described in male genitalia with lesions extending to the urethra and associated with

urethral and penile cancers while also reflecting a possibility of association with vagina cancer and pre-malignant vagina intra-epithelial neoplasia in females.^{1,5,4} This raises the need for proper evaluation and follow-up of patients with Lichen sclerosus especially in elderly where there is a high risk of transformation. Our second case was seen in an elderly woman, hence need for keen follow up.

Due to the recurrent nature of Lichen sclerosus and its frequently associated morbidities, issues with sexual difficulties often arises which can significantly impact on self-esteem and quality of life; both in pre-pubertal females as they mature into becoming sexually active and post-menopausal females who can be plagued by these sexual challenges and attendant discomfort in their genitalia. A study by Jablonowska et al in Italy showed significant impact on sexual quality of life, diminished work productivity and higher rate of depression among affected women.⁵ The association of LS with sexual life was also shown in a case report by Vettorazzi et al. in 2021.²⁴ This report was on a 69-year-old woman who had a history of intractable genital disease for over 20years.²⁴ Topical application of retinoids and testosterone, as well as the intramuscular injection of testosterone, were tried with limited success. The treatment process was further complicated by cases of dermatitis, sclerosis, and bleeding, in addition to the persistence of vulvar pruritus at age 52.²⁴ She has also experienced dyspareunia, low esteem with the aesthetics of her vulva, and pain after sexual intercourse.²⁴ This was corroborated in three studies by Haefner et.al in 2014, Cheng et.al in 2016 and Yildiz et.al in 2020, which showed the high incidence of sexual dysfunction in patients with Lichen sclerosus.²⁵⁻²⁷ However, history of sexual difficulty could not be ascertained in the elderly patient (Case 2) because she had not been sexually active for years prior to onset of symptoms.

Diagnosis of the condition is usually clinical; however, skin biopsy and histology may be needed to confirm the diagnosis and exclude possible differential diagnosis.^{4,10} Although some cases may be asymptomatic, treatment is often required to ease debilitating symptoms. Also, because of the likelihood of malignant transformation, lesions should be treated and patient followed up in case of possible transformation which can be seen in 4–6.7% of cases.⁴ Dermoscopy has recently been found to be a useful non-invasive diagnostic tool in Lichen sclerosus. Common dermoscopic patterns identified in Lichen sclerosus include follicular plugging with comedone –like openings and scales, rainbow pattern, grey-blue dots, usually with a characteristic peppered arrangement, corresponding to dermal melanophages, were also frequently seen. Others include presence of talangiectasias, whitish areas which represent areas of skin atrophy and fibrotic beams which are indications of a sclerosed dermis.^{4,8,28,29}

Treatment involves the use of topical clobetasol as its first line,² which is gradually tapered off with clinical improvement. This has been found to be effective in many cases as exemplified in the two cases presented. Recalcitrant forms of the disease with poor response to steroids can be managed by the use of calcineurin inhibitors, retinoids, oral and intralesional steroids.^{4, 8, 9, 30} Other treatment modalities are the use of fractional CO₂ laser, use of platelet rich plasma, photo-dynamic therapy and use of high frequency ultrasound. These have been tried with variable results. Cases of malignancies have been approached with surgery, radiotherapy and chemotherapy. Surgical maneuvers include surgical lysis of adhesions, perineoplasty and urethroplasty.⁴

In conclusion, lichen sclerosus is a chronic

inflammatory dermatosis which can affect all ages especially in extremes of life. Its occurrence has been under-reported in skin of colour and largely so in our environment. Its presence can be debilitating and impacts on quality of life of affected subjects; with a potential for malignant transformation and associated with depression and low productivity especially in females. This calls for a more holistic approach in its management with a multi-disciplinary approach to treatment and more reporting of cases especially in skin of colour which will aid disease surveillance and management.

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CONFLICT OF INTEREST:

The authors declare no conflict of interest.

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