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Case Report

Sign of Leser-Trélat: An association with benign swellings

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Abstract Sign of Leser-Trélat (SLT) is defined by sudden eruption of numerous seborrheic keratosis or rapid increase in the number of pre-existing seborrheic keratosis. It has been reported as a paraneoplastic manifestation in literature but its position as a paraneoplastic manifestation has both strong opponents and proponents. We hereby, report a case of a 49-year-old male who presented to us with SLT-like lesions in association with benign swellings of neck and scrotum.

Key words
Seborrheic keratosis, sign of Leser-Trélat, paraneoplastic, benign.

Introduction

Sign of Leser-Trélat (SLT), first described by Edmund Leser and Ulysse Trélat, is defined by the sudden eruption of numerous seborrheic keratosis or rapid increase in the number of pre-existing seborrheic keratosis,[1] preceding, revealing or following a malignancy. However, its position as a cutaneous paraneoplastic manifestation has always been questioned, as multiple seborrheic keratosis have been reported to occur in nonmalignant conditions like old age,[2,3] pregnancy,[4] HIV[5] and after receiving a heart transplant[6] etc. We, hereby, report a case of a middle aged man who presented to us with skin lesions simulating SLT along with neck and scrotal swellings, where all investigations failed to reveal any malignancy.

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Case report

A 49-year-old male, agriculturist by profession, came to us with complaints of development of a swelling on the neck and scrotum along with multiple, hyperpigmented cutaneous lesions.The cutaneous lesions were present for the last 18 years with rapid increase in number over the last two years. They were numerous, well-defined, hyperpigmented, hyperkeratotic, verruca-like papules and plaques varying in size from 1mm to 2 cm in diameter and were located on neck, upper chest and back arranged in a typical "rain drop" or "splash" pattern [**Figure 1(a)** and **1(a)**]. Lesions were occasionally pruritic and were all clinically consistent with seborrheic keratosis. The swelling on the neck was present for the last ten years and was gradually increasing in size, more so over the last two years. It measured 7x5 cm in size, was freely mobile, soft in consistency, fluctuant, non tender and the overlying skin was normal. The scrotal swelling was present for the last 18 years,was more on the right side than the left side and was gradually increasing in size for last four years. The scrotal

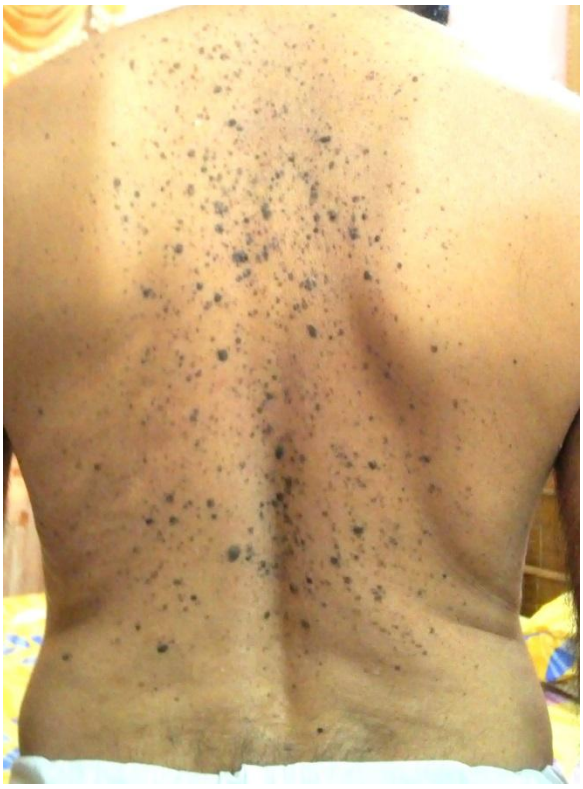

Figure 1 (a) Multiple seborrheic keratosis on the back of the patient. Raindrop / splash pattern of arrangement of lesions can be appreciated.

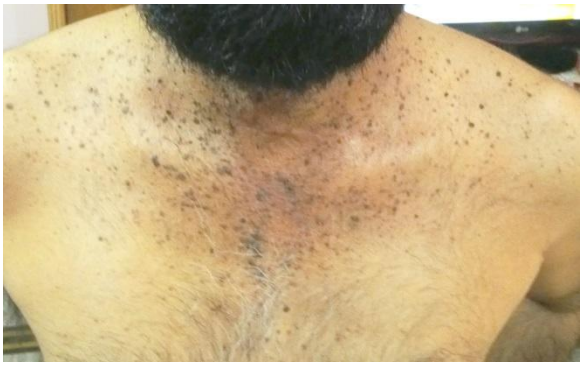

Figure 1 (b) Multiple seborrheic keratosis present on the neck and upper chest of the patient.

swelling was also nonpainful, nontender and the overlying skin was normal. The patient gave history of a renal transplant done 21 years ago for bilateral chronic renal failure due to chronic tubulointerstitial disease affecting both kidneys and since then he was on azathioprine125 mg BD and prednisolone 7.5 mg per day. There was no history of weight loss or any bowel or bladder complaint.

Keeping in mind the possibility of the Sign of Leser-Trélat and suspecting malignancy in this patient we got the patient investigated. General and physical examination were normal. Routine hematological, urine and stool examination and serum chemistries were normal and his HIV status was non reactive. Ultrasound of the scrotum showed bilateral hydrocele (right greater than left) whereas ultrasound of abdomen and pelvis was normal with no signs of lymphadenopathy. The neck mass was excised and sent for histopathological examination while for scrotal hydrocele eversion of sac was done and biopsy was taken from the sac. The neck mass on histopathology showed findings of epidermal inclusion cyst while no specific scrotal pathology could be appreciated in multiple sections of sac biopsy examined. Over a follow-up of six months, the patient is absolutely alright, has not shown any signs of malignancy or recurrence of swellings and the increase in number of seborrheic keratosis has come to a standstill.

Discussion

The SLT has been described with a wide variety of malignancies, especially adenocarcinomas, most of which arise in gastrointestinal tract[7,8] and also adenocarcinomas of other organs, and lymphoproliferative malignancies. It occurs in nonmalignant conditions like old age,[2,3] cytarabine therapy,[9] HIV, pregnancy etc. which condemns its status of a true paraneoplastic manifestation. Tumor-produced growth factors like epidermal growth factor and alpha-transforming growth factor have been hypothesized in the development of the seborrheic keratosis associated with malignancies.[10] Various patterns of SLT-like lesions have been reported like naeviform, dermatomal, blaschkoid, along skin cleavage lines and even as raindrops and streams.[2]

Heffernan and Khavari,[3] proposed that the raindrop pattern (linear, splayed, vertical distribution) represents a response to repeated overhead or near overhead sun exposure, hence the near vertical raindrop pattern. Our case was also consistent with these findings as he was an agriculturist by profession with long durations of sun exposure daily and also the localization of lesions to V area of chest along with photopigmentation further supports this theory.

We have reported this case because of the three questions it raises. First, the occurrence of SLT-like lesions in elderly has been considered to be normal, but the occurrence of lesions like SLT in a middle aged man without associated malignancy or any of the conditions reported so far was peculiar. Second, is there a role of long-term azathioprine therapy as a causative factor for inducing SLT like lesions similar to cytarabine?[9] Third, as no malignancy could be detected in this case, is it possible that growth factors responsible for benign swellings are sufficient enough to produce SLT-like lesions?

Conclusion

With this case being reported, the dilemma of SLT being a paraneoplastic sign remains unsolved. However, keeping in mind the malignant associations of SLT, both systemic and rarely development of malignancy within seborrheic keratosis themselves,[11] any patient presenting with SLT-like lesions should be thoroughly investigated to rule out any malignancy and followed up regularly.

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