CASE REPORT

CUTANEOUS MANIFESTATIONS OF MIGRAINE: FIRST OF ITS KIND, TWO CASE REPORTS FROM PAKISTAN

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Migraine is a common clinical entity; however, its dermatological manifestations are rarely reported. We report two young Pakistani female patients with asymptomatic, linear and round erythematous non blanchable patches on their forehead that were temporally associated with their migraine attacks. The local and systemic examination as well as extensive investigations, of both the patients, were all normal. The lesion resolved within a few days following abortion of migraine attacks in these patients. With Pathophysiology of cutaneous manifestations of migraine is still unknown due to their rare presentation, research is required to establish their significance in the long-term outcome of migraine.

Keywords: Migraine; Red dot syndrome; Cutaneous manifestations of migraine

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INTRODUCTION

Migraine is a well explored neurological entity, characterized by episodic unilateral or bilateral attacks of headaches with signs and symptoms of aura such as dizziness, nausea and photophobia with a diagnostic criterion that helps in its diagnosis.[1] There are other manifestations, reported over the course of time; rare entities like "primary erythromelalgia in facial region" and "red ear syndrome" found in association with migraine have been reported[2], however rest of the cutaneous manifestations of migraine are reported sparsely[3,4]. We report two Pakistani young women who presented separately over a period of six months, with asymptomatic erythematous linear and round patches respectively, on their foreheads at the time of migraine attacks. The presentations of these lesions were directly related to the severity of migraine attacks in both the patients.

CASE-1

A 37 years of age dextromanual female homemaker, married, two kids for eight years with no other comorbid, presented to dermatology outpatient department with complaints of multiple episodic red linear streaks on her forehead appearing for four years off and on. These lesions were asymptomatic however they were associated with severe unilateral throbbing headache that upon further inquiry, fulfilled the criteria of migraine without aura. She had been suffering from these episodic attacks of headache, for the past four years without being labelled as a case of migraine by any doctor. She used oral non-steroidal anti-inflammatory medications to relieve her attacks. There were no other local or systemic complaints or any indicators of focal neurological deficit. On examination, her vitals were found to be within normal range, i.e., temperature 98.6 °F, pulse rate 90 beats/min, breathing rate 15 breaths/min and blood pressure was 135/85 mmHg. Her general physical examination was unremarkable while cutaneous examination revealed multiple erythematous, asymptomatic, linear non blanchable, vertical patches on her forehead, the longest streak being 6 cm in size while the smallest, 2.5 cm in size. She refused to get photographed during the initial attack due to cultural preferences however later agreed when she had another episode of migraine with similar skin manifestations, where the erythematous patch was again linear but single, measuring around 7 cm in length (Figure-1). Rest of her systemic examination including cardiovascular, neurological and abdominal examination was unremarkable.

The patient was reassured and placed in a dark room. She was given oral NSAID (Ibuprofen) for headache. Her baseline investigations including blood complete picture, liver & renal function tests, electrocardiogram and urine routine report were sent which were later found to be normal. Her MRI brain study was also found to be normal. After thorough counselling and consent, a punch biopsy of the cutaneous lesion was taken and sent for histopathological analysis which showed epidermis hyperkeratosis. Subcutaneous tissue showed red blood cells extravasation. Focal crusted vessels were also seen. There were no lymphocytes/ eosinophils, extravasation and the collagen bundles were found be normal looking (Figure-2,3,4). The headache subsided

considerably in the ensuing hours. The cutaneous markings, however persisted longer, gradually fading in colour and length until completely resolved in the next 36 hours in all of her clinical presentations. After 2 weeks, the patient reported with complaints of headache, of lesser intensity with proportionately less marked forehead markings than the initial attack. Following oral medications which abolished the migraine attack, the cutaneous markings resolved on second day of resolution of headache. With all her investigations being normal, the patient was referred to neurological OPD to start her on migraine prophylaxis treatment. She was counselled about the rarity and seemingly benign nature of these lesions. She agreed to be followed up on term basis and remain in touch with dermatologist reporting this case.

CASE-2

A 27 years of age dextromanual female student, known case of migraine, with no other comorbid, presented to dermatology outpatient department with complaints of multiple episodic discoid erythematous lesions on her forehead (Figure-5) appearing for three years off and on. Like the first case, these lesions were asymptomatic associated with severe unilateral throbbing headache that fulfilled the criteria of migraine without aura. She used abortive therapies for her migraine attacks as she didn't meet the criteria to put her on migraine prophylaxis and ignored her skin markings until recently as she planned to get married soon. There were no other local or systemic complaints or any indicators of focal neurological deficit. Her vitals, general physical and systemic examination were all unremarkable while cutaneous examination revealed four asymptomatic erythematous discoid patches with surrounding oedema.

The two larger patches measuring 2x3 cm in size while the other two were much smaller and fainter. The baseline investigations including blood complete picture, liver & renal function tests, electrocardiogram and urine routine and MRI brain study, were all normal like the first patient. She refused to get biopsied despite counselling. The cutaneous markings, followed a course, similar to the first case where they persisted longer than the headache and gradually faded in colour and size until completely resolution in the next two days. With all her investigations being normal, the patient was counselled in detail about the rarity and seemingly benign nature of these markings and also advised a close follow-up in dermatology and neurology outpatient departments.

The differential diagnosis in both cases included migraine, primary erythromelalgia presenting as migraine, *morphea en coup de sabre*, a space occupying lesion and vascular tumour. The history, examination and investigations in both cases ruled out other differentials. Both the fulfilled the diagnostic criteria of migraine without aura and hence were diagnosed as cases of migraine with cutaneous manifestations.

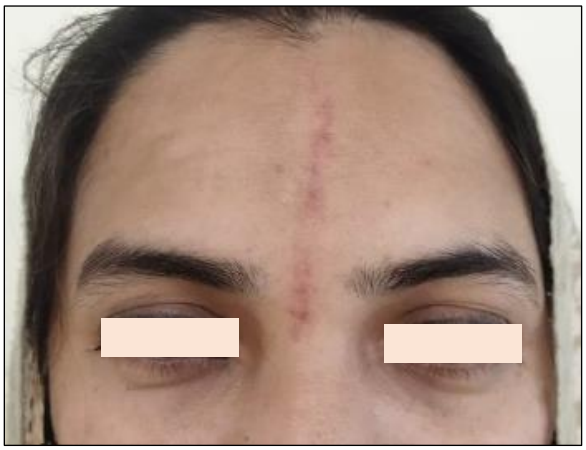

Figure-1: Patient #1, presenting with episodic red linear streaks on her forehead appearing for four years off and on.

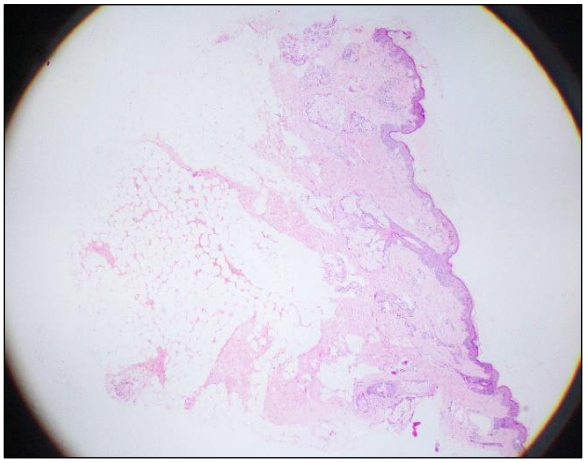

Figure-2

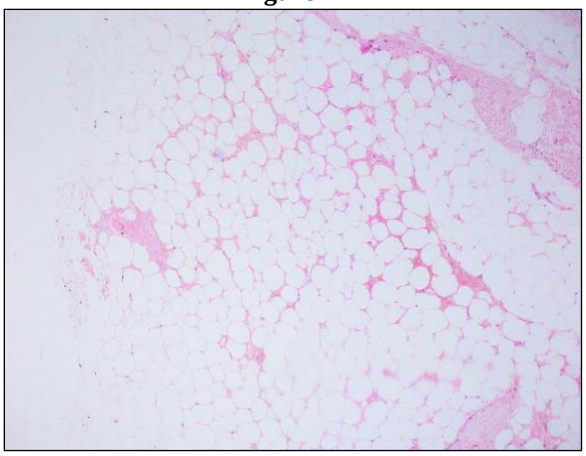

Figure-2

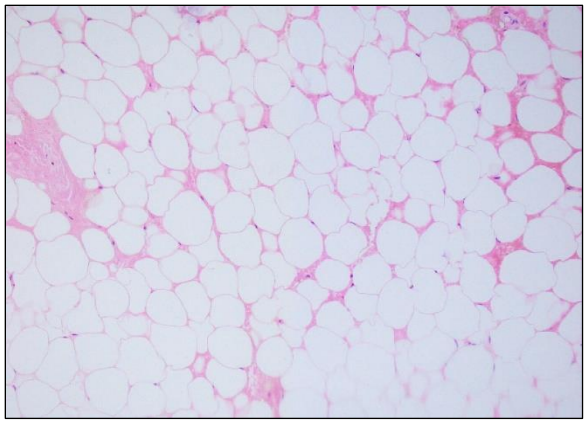

Figure-2,3,4: Biopsy of lesions from patient #1, showing subcutaneous tissue with red blood cells extravasation. Focal crusted vessels are also seen.

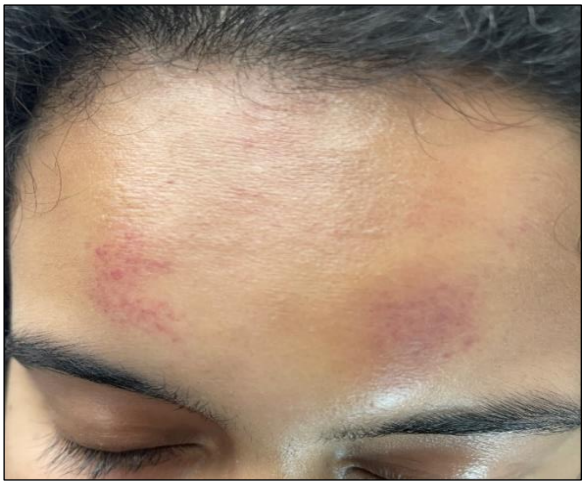

Figure-5: Patient #2 with multiple episodic discoid erythematous lesions on her forehead.

DISCUSSION

The cutaneous manifestations of migraine have been reported via only a few case reports.[3,4] While differentials like *morphea en coup de sabre* and vascular tumours were ruled out with cutaneous biopsy, the CT brain ruled out any intracranial SOL. The important differential of Primary Facial Erythromelalgia of Migraine[2] was rule out with history and examination as the lesions, although associated with migraine, were completely asymptomatic, had no known triggering/aggravating factors and would resolve completely, only following the resolution of migraine attacks. To the best our knowledge this is the first instance where two female patients of migraine with such extensive cutaneous involvement are reported. These are also the first case reports of such nature from Pakistan.

Interestingly, of the total seven cases reported so far, three patients are of Pakistani origin. The cutaneous manifestations in our first case are similar in distribution and behaviour to an earlier report of a UAE based male of Pakistani origin, not limited to V1 distribution of trigeminal nerve.[3] The earliest reports have described the cutaneous manifestations of migraine as a red dot in a female of Turkish and European origin, both in the distribution of the frontal branch of trigeminal nerve.[3-5] The Biopsy findings in our sample corroborate the previous suggestions regarding the nature of these cutaneous manifestations being vascular in origin.[4]

CONCLUSION

The cutaneous manifestations of migraine are rare and need further exploration and research regarding their pathophysiology and association with migraine severity and prognosis. Based on the so far higher frequency of these cutaneous manifestations of migraine in eastern Mediterranean ethnicities in general and Pakistani population in particular, we hypothesize that such cutaneous findings may have a predilection to a specific ethno-geographic distribution. There may also be a predilection of such findings in female patients. Clinicians need to be aware of such manifestations and may follow such patients on long term basis to understand the outcome of their illness.

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