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CASE REPORT

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Palmar papules as a manifestation of cutaneous oxalosis in primary hyperoxaluria: A case report and review of the literature

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Key Clinical Message

Cutaneous oxalosis is a rare manifestation of systemic oxalosis, typically associated with primary or secondary hyperoxaluria. We present a rare case of a 23-year-old female diagnosed with primary hyperoxaluria and end-stage renal disease, who presented with papules on the palms without any vascular complications. The skin can be affected by oxalate deposition, resulting in various manifestations such as vascular complications or calcified nodules. In our case, the patient had primary hyperoxaluria and end-stage renal disease but exhibited atypical features of cutaneous oxalosis. Histopathology confirmed the presence of oxalate crystals in the dermis, subcutis, and medium-sized arteries. The mechanism of oxalate deposition in this case remains unclear. This case underscores the importance of considering cutaneous oxalosis in the differential diagnosis of patients with renal failure and skin lesions, and highlights the variability of clinical presentations in primary hyperoxaluria.

KEYWORDS

cutaneous oxalosis, oxalosis, palmar papules, primary hyperoxaluria

1 | INTRODUCTION

The term "oxalosis" refers to the systemic buildup of calcium oxalate, the insoluble salt of oxalic acid, outside of the urinary system. Primary and secondary hyperoxalurias are the pathologic disease processes in charge of causing systemic oxalosis. The kidneys, bone, heart, blood arteries, and skin are major sites of oxalate accumulation that result in illness.[1] Primary hyperoxalurias may be characterized by vascular deposition of skin manifestations that produce oxalate, such as livedo reticularis, acrocyanosis, peripheral gangrene, and ulcerations.[2] Skin symptoms, on the contrary, are uncommon in individuals who develop an oxalosis caused by renal insufficiency and, when they do, they are the consequence of extravascular deposition, resulting in calcified nodules and miliary papules.[3] Globally, an estimated genetic prevalence was 1 in 58,000 people who suffer with primary hyperoxaluria. With Type 1 accounting for around 80% of cases.[4] PH type I is caused by mutations in the AGXT gene, which encodes the liver-specific enzyme alanine-glyoxylate aminotransferase (AGT), AGT is responsible for converting glyoxylate to glycine, thereby preventing the formation of oxalate.[5] PH type I patients have a high risk of early end stage kidney

failure, which may then lead to systemic oxalate deposition.[6] Here we report a case of a 23-year-old female patient with a cutaneous oxalosis without livedo reticularis, acrocyanosis and ulceration, and it could be a very rare case in medical literature.

2 | CASE PRESENTATION

A 23-year-old female came from department of renal diseases diagnosed with primary hyperoxaluria with end-stage renal disease undergoing dialysis 2 years ago three times a week 4 h a day. The patient needs a liver and kidney transplant and presented to the dermatology clinic with a consultation for papules on palms by the nephrologist for his suspicion of warts (Figure 1A–C). And this is a containdication for kidney and liver transplantation. Returning to the patient's family story, there is a deceased brother and father with the same disease. The patient recalls being healthy until her early 20s when she experienced two episodes of nephrolithiasis. Approximately a year and a half later, she noticed the development of the papules on palms. Dermatological examination was significant for this woman. However, based on her initial history and physical examination, the differential diagnosis was primary hyperoxaluria versus calciphylaxis. Histologic examination of biopsy showed radially arranged yellow-brown rhomboid crystals surrounded by histiocytes in the subcutis and reticular dermis (Figure 2A,B). Findings from the patient's laboratory workup notable for both mild hypocalcemia (8.6 mg/dL) as well as normal platelet count and glucose level. Based on her laboratory findings and biopsy results and given her history, a diagnosis of renal insufficiency in the setting of primary oxalosis.

3 | DISCUSSION

Cutaneous oxalosis is a rare manifestation of systemic oxalosis, which is usually associated with primary or secondary hyperoxaluria. Primary hyperoxaluria is a genetic disorder that causes overproduction of oxalate by the liver, leading to excessive urinary excretion and deposition of oxalate in various tissues. Secondary hyperoxaluria is caused by increased intestinal absorption or dietary intake of oxalate or its precursors. Both types of hyperoxaluria can result in kidney stones, renal failure, and systemic oxalosis.[7] The skin is one of the organs that can be affected by oxalate deposition, especially in patients with primary hyperoxaluria and renal failure. The most common cutaneous manifestations of oxalosis are vascular complications, such as livedo reticularis, acrocyanosis, peripheral gangrene, and ulcerations. These are caused by the occlusion of small blood vessels by oxalate crystals, leading to ischemia and necrosis of the skin.[7,8]

The histopathology confirmed the presence of oxalate crystals in the dermis, subcutis, and medium-sized arteries, but there was no evidence of vascular occlusion or ischemia. The mechanism of oxalate deposition in this

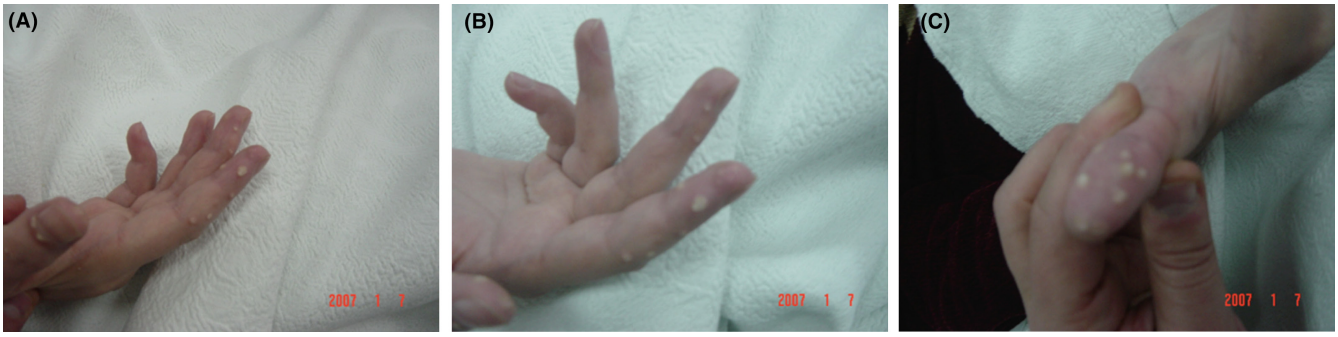

FIGURE 1 (A) shows the papules on palms and hopefulness. (B) shows the papules on palms and hopefulness. (C) shows the papules on palms and hopefulness.

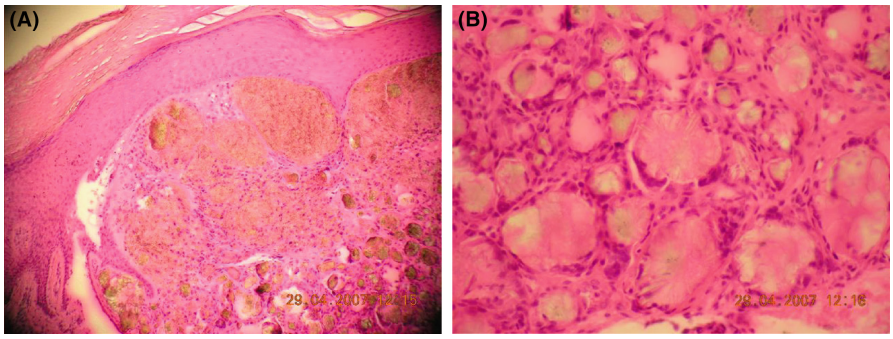

FIGURE 2 (A) shows Histologic examination of biopsy of primary hyperoxaluria. (B) shows Histologic examination of biopsy of primary hyperoxaluria.

case is unclear, but it may be related to local factors such as trauma, inflammation, or infection that facilitate the crystallization of oxalate in the skin.[7] The diagnosis of cutaneous oxalosis can be challenging, as it can mimic other conditions such as calciphylaxis, nephrogenic systemic fibrosis, or warts. The clinical history, laboratory tests, and stone analysis can provide clues to the underlying cause of hyperoxaluria and oxalosis. However, the definitive diagnosis requires a skin biopsy that shows characteristic birefringent crystals under polarized light microscopy.[2]

Our case differs from previous reports of cutaneous oxalosis in several aspects. First, most of the reported cases of cutaneous oxalosis were due to secondary hyperoxaluria, whereas our case had PH1, which is a rare and severe form of the disease.[9] Second, most of the reported cases of cutaneous oxalosis had vascular complications, such as livedo reticularis, ulceration, or gangrene, whereas our case had only papules on the palms without any signs of vascular occlusion or ischemia.[10] Third, most of the reported cases of cutaneous oxalosis had oxalate deposits in the small vessels of the dermis, whereas our case had oxalate deposits in the medium-sized arteries of the subcutis as well.[10] These differences suggest that cutaneous oxalosis has a heterogeneous presentation and may involve different mechanisms of oxalate deposition and tissue damage.

The treatment of cutaneous oxalosis depends on the type and severity of hyperoxaluria and renal function. The main goals are to reduce the production and excretion of oxalate, prevent stone formation and infection, preserve renal function, and manage pain and complications. The treatment options include dietary modification, hydration, medication (such as pyridoxine, citrate, or phosphate), dialysis, or transplantation (kidney or combined liver-kidney).[2,7]

In conclusion, we report a rare case of cutaneous oxalosis due to primary hyperoxaluria and end-stage renal disease that presented with papules on the palms without any vascular complications. This case highlights the variability of clinical presentations in primary hyperoxaluria and the importance of skin biopsy for diagnosis. Cutaneous oxalosis should be considered in the differential diagnosis of patients with renal failure and skin lesions.

AUTHOR CONTRIBUTIONS

Hadi Alabdullah: Writing – original draft; writing – review and editing. **Jameel Soqia:** Writing – original draft; writing – review and editing. **Thaer Douri:** Supervision.

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Not applicable.

DATA AVAILABILITY STATEMENT

Not applicable.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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