

# Acquired Kyrle's disease at the site of healed herpes zoster

Mohammed El-Sayed Hanafy Khalel, Amna R. Al Mula, Adel H. Al-Abdul Razzaq, Nabeel Najem, Salhamoud Abd El-Fatah Saleh\*, Omar Shamsaldeen\*\*

Department of Dermatology, Adan Hospital, Kuwait

\*Department of Medicine, Adan Hospital, Kuwait

\*\*Department of Dermatology, Farwaniya Hospital, Kuwait.

**Abstract** Kyrle's disease is a rare variant of primary perforating dermatosis. We reported a case of Kyrle's disease involving the site of healed herpes zoster. The clinical picture shows follicular and interfollicular hyperkeratotic horn-like plugs at the site of the resolved herpes zoster. It is thought to be an isotopic response that describes the occurrence of a new skin disorder at the site of another lesion that has already healed and is unrelated.

**Key words**

Kyrle's disease, herpes zoster, Isotopic phenomenon.

## Introduction

Kyrle in 1916 described a dermatosis which he named 'hyperkeratosis follicularis et parafollicularis in cutem penetrans'.<sup>1</sup>

The primary lesion of Kyrle's disease (KD) is a reddish-brown papule or nodule with central keratin plugs. The lesions may be asymptomatic, or itchy with underlying systemic illness such as diabetes mellitus, renal failure or hepatic failure.<sup>1,2</sup> The lesions of KD may be follicular or parafollicular. Characteristically, the disease does not involve the mucous membranes and palmoplantar surface.<sup>3</sup> Several authors have reported the involvement of mucous membranes by KD e.g. conjunctiva and buccal mucosa.<sup>4</sup> Histopathologically a keratotic plug fills an epithelial invagination. Parakeratosis is present in parts of the plug that sometimes penetrates in

the dermis.<sup>5</sup> Herein, we report a case of KD arising at the site of healed herpes zoster.

## Case report

A 27-year-old Bengali male was diagnosed as herpes zoster on the upper left trunk. Two months later, he presented with a zosteriform, brown, papular and mild itchy eruption at the site of the resolved herpes zoster.

The physical examination of the skin lesions revealed multiple brown papules with central umbilication that contained firmly adherent keratotic plugs on the upper left trunk, **Figures 1, 2 and 3**. There was no preceding history of trauma and no signs or symptoms of any systemic disease, metabolic condition such as diabetes mellitus, chronic renal failure or hepatic failure. He had no history of a similar skin eruption in his family. Laboratory investigations revealed normal blood sugar, renal and liver function tests. The biopsy specimen showed dilated hair follicle filled with cornified plug

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**Address for correspondence**

Dr. Mohammed El-Sayed Hanafy Khalel, MD,  
Senior Specialist,  
Department of Dermatology,  
Adan Hospital, Kuwait  
E mail: mohammedhanafy\_1957@hotmail.com



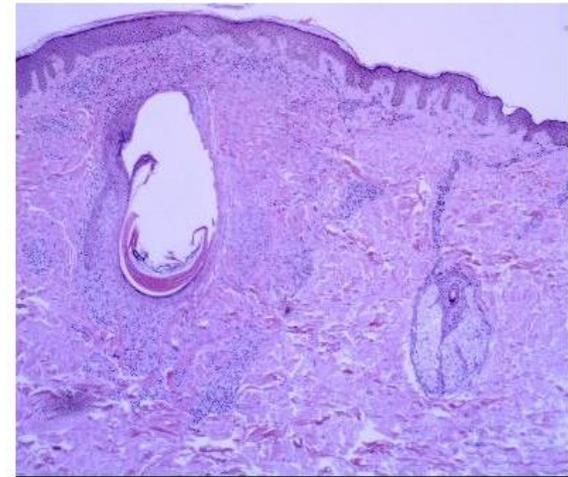
**Figure 1** Zosteriform, brown, papules erupted at the site of resolved herpes zoster.



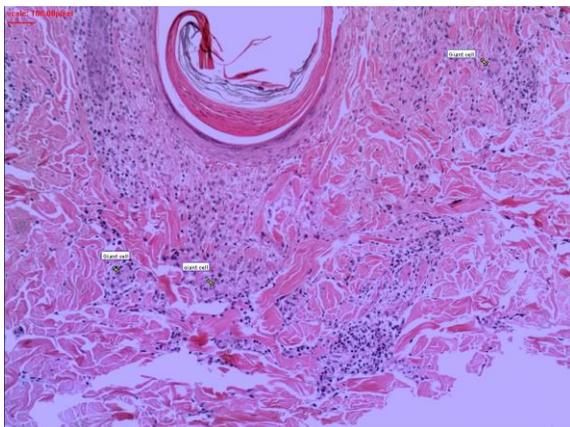
**Figure 2** High power magnification revealed umbilicated papules with firmly adherent keratotic plug.



**Figure 3** Zosteriform, brown, papules on the back.



**Figure 4** Skin biopsy showing dilated hair follicle, filled with cornified plug with parakeratosis. Mild irregular epithelial hyperplasia and marked lymphohistiocytic infiltrate in the dermis surrounding the hair follicle is noted. Macrophages with dark brown pigment are also scattered [H&E x 40].



**Figure 5** Examination under high power revealed giant cells surround hair follicle [H&E x100].

along with parakeratosis and basophilic debris. No elastic or collagen fibers were identified by Verhoeff's elastic and Masson trichrome stain.

Mild irregular epithelial hyperplasia was noted and a marked lymphohistiocytic infiltrate surrounded the hair follicles in the dermis. Giant cells were also scattered in the dermis. These findings are consistent with KD (**Figures 4** and **5**).

## Discussion

Many cutaneous diseases that occur at the site of

resolved herpes zoster have been reported.<sup>6</sup> These include granuloma annulare, sarcoidal granuloma, tuberculoid granuloma, lymphoma and Kaposi's sarcoma. There have been, however, only three reports of reactive perforating collagenosis at the healed site of herpes zoster.<sup>7,8,9</sup> We report a case of acquired KD that developed at the site of a previous herpes zoster skin eruption in a 27-year-old Bengali male. This is thought to be an isotopic response that describes the occurrence of a new skin disorder at the site of another lesion that has already healed and is unrelated to primary dermatosis.<sup>6</sup> KD is one of the perforating skin diseases that represents a heterogeneous group of disorders characterized by transepithelial elimination of dermal structures.<sup>10</sup> Primary perforating disorders include Kyrle's disease, elastosis perforans serpiginosa, perforating folliculitis and reactive perforating collagenosis.<sup>11</sup> These need to be distinguished from the secondary perforating dermatoses due to metabolic conditions such as uremia especially in patients on dialysis, diabetes mellitus, hepatic failure or as paraneoplastic syndrome in multiple myeloma.<sup>12</sup> KD has been also reported with other conditions including tuberculosis, pulmonary aspergillosis, scabies, atopic dermatitis, AIDS, neurodermatitis and malignant and endocrinological disorders.<sup>13</sup> KD usually appears between 30 and 50 years of age, however, there has been a case report of this disease in a 75-year-old patient<sup>14</sup> as well as in a 5-year-old child.<sup>15</sup> KD was found in a family of three generations, suggesting an autosomal dominant mode of heredity.<sup>5</sup> However, in more recent publications, KD is considered as an autosomal recessive genodermatosis.<sup>16</sup> In our patient no hereditary association was confirmed. KD occurs commonly in the setting of diabetes mellitus,<sup>17</sup> chronic renal failure,<sup>18</sup> hepatic insufficiency<sup>16</sup> or hyperlipoproteinemia<sup>19</sup> and

rarely may be paraneoplastic disease.<sup>12</sup> The exact etiopathogenesis of KD remains unknown. In 1961, it was thought that arsenic might be related to the etiology of this disease.<sup>20</sup> Nowadays, several hypotheses have been proposed for pathogenesis of KD including diabetes mellitus related microangiopathy, microtrauma due to chronic pruritus and abnormalities of collagen, elastin and vitamin A or D metabolism in patients with renal disease.<sup>21</sup> It is suggested that one of the extracellular matrix protein, fibronectin could be involved in the pathophysiological mechanism in KD as well as perforating dermatoses.<sup>22</sup> The role of infectious agents, probably anaerobic bacteria, in the pathogenesis of KD is supported by response with clindamycin<sup>23</sup> and with metronidazole.<sup>24</sup>

An isotopic response describes the occurrence of a new skin disorder at the site of another lesion that has already healed and is unrelated.<sup>6</sup> The preceding diseases is frequently herpes zoster. The pathogenesis of the cutaneous reactions that follow resolved lesions of herpes zoster remains unclear. Immunologic factors, including exaggerated hypersensitivity tissue antigens in the original viral infection, may be involved in the pathogenesis of the second disease.<sup>6</sup>

To our best knowledge this is the first case of KD which occurred at the site of healed herpes zoster to be reported in English literature.

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