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A Bullous Pilomatricoma Developed after Hepatitis A Vaccination

The Editor,

Sir,

Pilomatricomas originate from hair matrix cells and usually appear as firm, solitary and asymptomatic nodules beneath the skin. These tumours occur mostly in children. They are generally located on the face and neck (1). Bullous pilomatricoma is an uncommon lesion and only few cases of this variant have been reported in the literature (1, 2).

We report a 7-year old girl with a pilomatricoma showing bullous appearance. The patient suffered from a dome shaped, bullous mass on the left arm. The 1.5 × 1.5 cm lesion presented one month previously and progressively enlarged. In the history of the patient, there was a hepatitis A vaccination to the same area with the bullous lesion, about four months earlier. Two days after the vaccination, severe inflammation occured in the same region. The patient did not have any family history of such or mechanical trauma. On dermatologic examination, a dome shaped, red-brown coloured, semi-translucent bullae was noted (Fig. 1A).

The bullae collapsed inward with palpation and a firm, small, painless mass was felt at the bottom of the lesion. When the lesion was pressed with the tip of a pen, it had a wrinkled-atrophic appearance (Fig. 1B). The other physical and systemic examination findings were normal and there was no lymphadenopathy. Routine haematological and biochemical examinations of the patient were within the normal limits. With dermatoscopy, red-coloured tortuous small vessels on an irregular white opacity, which settled on a livid-red background, were observed (Fig. 1C).

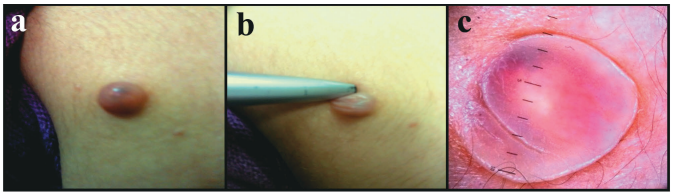

Fig. 1: (A) Clinical appearance of the bullous pilomatricoma; (B) pressed and wrinkled appearance of the lesion and (C) dermatoscopic appearance of the lesion.

Needle aspiration material of the bullae was haemorrhagic and its microbiological culture was sterile. With these findings, the diagnosis of the lesion was made as a "bullous pilomatricoma". The mass was totally removed with surgical excision. On histopathological examination, the tumour nests were composed of eosinophilic shadow cells, basophilic cells and surrounded by a fibrous capsule in the deep dermis. Calcium salt depositions were observed in the tumour mass. In the superficial dermis, marked lymphoedema and increased numbers of dilated lymphatics filled with eosinophilic lymph fluid were observed (Fig. 2).

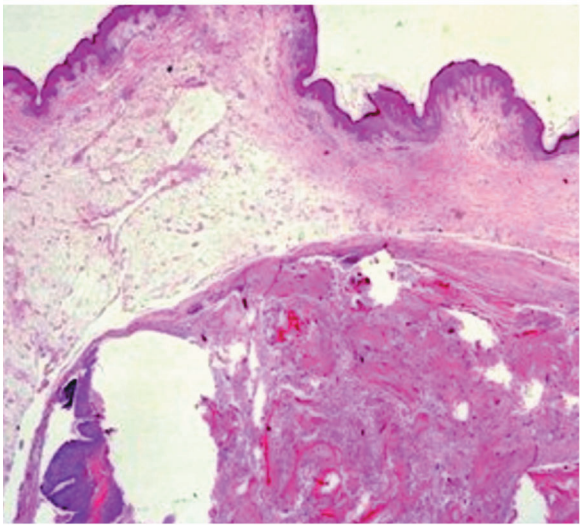

Fig. 2: Histopathologic appearance of the lesion (HE × 40).

Bullous pilomatricoma is a very rare form of pilomatricoma (3). Their incidence is between 3% and 6% (1). Although pilomatricomas can be associated with other genetic disorders such as myotonic dystrophy and Gardner's syndrome, the bullous variant is not associated with these syndromes (2). The bullous lesions are located mostly in the shoulder and upper extremity and predominantly in females. They are usually asymptomatic lesions (1, 2). Clinical characteristics reported include semi-transparent, erythematous, bluish or skin-coloured, heavily folded or striae-like, flaccid blisters overlying a solitary, firm-to-hard nodule (2, 4). Mechanical irritations such as continuous mechanical stimulation, scratching and pinching trauma and continuous pressure play an important role in the development of bullous pilomatricomas (2, 5).

The common dermatoscopic findings of bullous pilomatricomas are reddish homogenous areas, irregular white structures and hairpin-like atypical vessels (6). In the histopathology of pilomatricomas can be seen eosinophilic shadow cells, basophilic cells, foreign body cells, calcified focus or even ossification (2). In a bullous pilomatricoma, in addition to these findings, lymphoedema and dilated lymphatic vessels in the superficial dermis were found in most cases (1–3). These lymphatic findings have been described as common pat-

hologic features (2). The skin overlying pilomatricoma sometimes can be atrophic. It is supposed that this atrophic appearance is related to the loss of elastic fibres, and dermal oedema (1). In the aetiopathogenesis, it is suggested that the cause of the bullous appearence of the tumour is obstruction of lymphatics, and congestion by lymphatic fluid (1, 2, 5).

In addition, some authors suggest that the tumour cells can produce elastinolytic enzymes. These lytic products cause disruption of collagen fibres and destruction and dilation of lymphatic vessels (2, 5). After these processes, the dermis is filled with lymphatic fluid (2). Since the lesions do not regress spontaneously, treatment is surgical (1). Our patient had all the features of a typical bullous pilomatricoma. Additionally, in the history of the patient, there was hepatitis A vaccination to the same region with the bullous lesion and thereafter a severe inflammation developed in the vaccination area. Therefore, we thought that both the pricking trauma of vaccination and inflammation caused by the vaccine might be the trigger of the aforementioned mechanisms. To our best knowledge, the case is the first bullous pilomatricoma which developed after the hepatitis A vaccination. Our case is being reported to draw attention to this rare entity and a probable aetiopathogenetic association with hepatitis A vaccine and bullous pilomatricoma.

Keywords: Bullous, hepatitis A, pilomatricoma, tumour, vaccination

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DOI: 10.7727/wimj.2013.304

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Can Isotretinoin Induce Articular Symptoms in SAPHO Syndrome?

The Editor,

Sir,

A 28-year old man visited his doctor because of hip pain (worse on the left side) and low back pain (worse with immobility/at night and better with activity) for the last 10 days. He stated that he had used oral isotretinoin for acneiform lesions for two weeks, and his complaints had ensued thereafter. He added that he had experienced a similar history with isotretinoin treatment when he was 20 years old as well. On physical examination, bilateral hip joint movements and lumbar flexion were painful. He also had limited range of motion in his left hip. Sacroiliac joints, sternum and sternoclavicular joints were painful with palpation. Gaenslen's and Mennel tests were positive, bilaterally. He had antalgic gait on the left side. He had severe nodulocystic acne with abcesses on his face, neck and back. C-reactive protein level was 1.87 mg/dL (N: 0–0.8 mg/dL). Laboratory tests including erythrocyte sedimentation rate, liver and kidney function tests and Brucella Rose Bengal test were all within normal limits. Radiographs were unremarkable. Magnetic resonance imaging (MRI) showed bilateral sacroiliitis (Fig. 1). Bone scintigraphy was consistent with bull's head sign (Fig. 2).

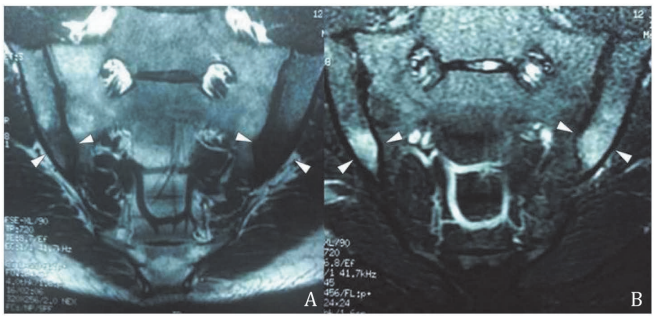

Fig. 1: Magnetic resonance imaging demonstrating bilateral sacroiliitis (white arrowhead) on T1-weighted **(A)** and T-2 weighted **(B)** axial views.

Eventually, the patient was diagnosed with SAPHO (synovitis, acne, pustulosis, hyperostosis and osteitis) syndrome possibly induced by isotretinoin treatment. Isotretinoin was stopped and the patient was treated with indomethacin 75 mg/d. There was marked improvement in his complaints (except for acne) in the second month of follow-up.

SAPHO syndrome is a chronic inflammatory disorder with findings of synovitis, acne, pustulosis, hyperostosis and