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CASE REPORT

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Bullous Pemphigoid Triggered by Artificial Hip Made of Titanium Alloy: a Case Report and Review of Triggers for Bullous Pemphigoid

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Abstract:

Bullous pemphigoid (BP) is one of the most common autoimmune blistering diseases. Here, we report an old woman presented with a 2-month history of bullous lesions located just over the skin of the right thigh and buttock where the orthopedics operation was performed using artificial hip made of titanium alloy and a twenty days history of similar lesions involving the rest of the body gradually.

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Running title:
BP triggered by artificial hip.

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Introduction

Bullous pemphigoid (BP) is one of the most common autoimmune blistering diseases. Most cases develop in the absence of an identifiable trigger, and generally occur in elderly patients [1]. Although BP is usually idiopathic, some cases induced by chemical and physical triggers as well as medications have been reported [2,3]. We report a case of typical BP which was strongly suspected to have been triggered by the artificial hip made of titanium alloy. To our knowledge, this is the first reported case of BP by the artificial hip. Artificial hip should be added to the list of possible triggers for BP and the diagnosis should be considered if blistering develops following hip replacement surgery.

Case Report

We report a 77-year-old woman presented with a 2-month history of bullous lesions located just over the skin of the right thigh and buttock where the orthopedics operation was performed and a twenty days history of similar lesions involving the rest of the body gradually before the patient was referred to our hospital.

At the beginning of the episodes, she experienced a sudden onset of a femoral neck fracture caused by an accidental fall in May 2011. Then the patient was referred to hospitalization for orthopedics operation at a local hospital. Only seven days after undergoing hip arthroplasty made of titanium alloy, the patient reported severe pruritus and rapid onset of erythema and blistering located just over the skin of the right thigh and buttock where the orthopedics operation was performed. At first presentation, the dermatologist in that hospital diagnosed this patient with contact dermatitis and initiated oral antihistamine, topical application of drug as the main treatment. However, despite the treatment, the patient experienced the recurrent attacks of vesicles, bullae and blood blister for about forty days. More seriously, similar lesions then expanded to the rest of the body gradually for twenty days.

In July 2011, the patient was admitted to our ward due to recurrent episodes of skin lesions. On physical examination, multiple tense vesicles, bullae and blood blister were seen over the extremities and trunk with a background of erythema (Figure 1A and B). In addition, Nikolskiy's sign was negative. A clinical diagnosis of BP was considered based on the clinical manifestation. The right hip prosthesis made of titanium alloy can be seen by the computed tomography scanning Figure 1C). The patient had a history of penicillin allergy, 17-year-history of hypertension and no personal history or family history of BP. She took metoprolol succinate sustained-release tablets to lower blood pressure for more than 8 years. The biopsy specimen taken from a recently developed intact vesicle on the patient's left arm shows

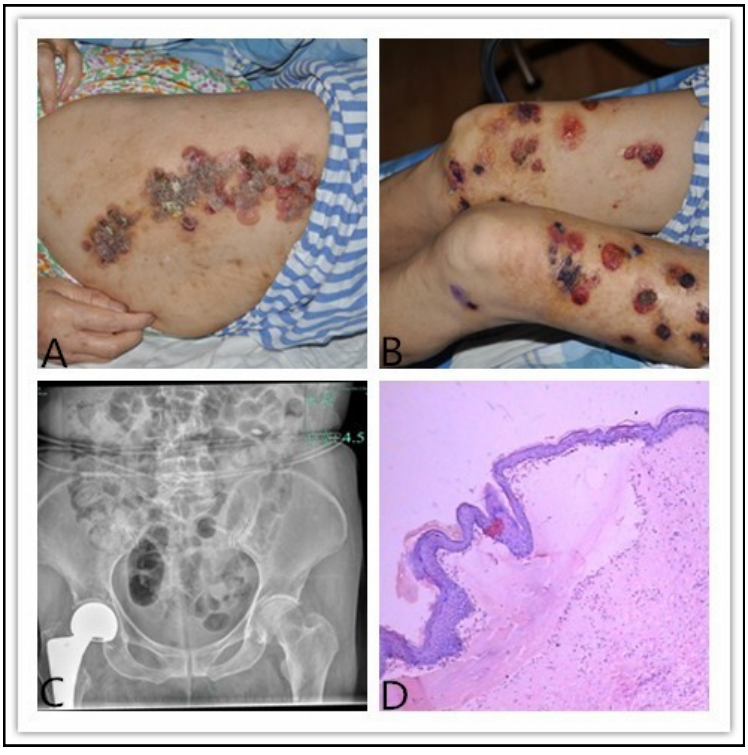

Figure 1. (A) Erythema and blisters on the skin of the right thigh. **(B)** Multiple tense vesicles, bullae and blood blister on the legs on a background of erythema. **(C)** The right hip prosthesis made of titanium alloy can be seen by the computed tomography scanning. **(D)** Histopathology revealed a subepidermal blister (haematoxylin and eosin staining, original magnification x 100).

a subepidermal blister, filled with an inflammatory cell infiltration consisting of numerous eosinophils, neutrophils, and a small number of lymphocytes (Figure 1D). Direct immunofluorescence showed IgG and C3 at the dermo-epidermal junction. Moreover, we detected the presence of circulating antibodies against BP180 in this patient . All the clinical manifestations and laboratory examinations confirmed the final diagnoses of BP. The patient was treated with high-dose intravenous methylprednisolone (60mg once daily for one week) and intravenous immunoglobulin (IVIG, 0.4g/kg/d for 5d). Then oral prednisone 50 mg daily was continued and the lesions had completely resolved within 5 weeks.

Discussion

BP is one of the most common autoimmune blistering diseases. Many factors may be responsible for the onset of BP including drug intake, physical agents, and viral infections [4]. Medications are the most common cause of development for BP. Both systemic and topical drugs have been reported to be triggers in the development of BP such as iodine and etanercept [1], dipeptidyl peptidase-IV inhibitors plus metformin [5], loop diuretics [6], ophthalmic preparations [7]. In addition, thermal burns [8-13] and boiling water burn [2] can induce BP. Scabies cannot be ruled out of the list of probabilities, scabies infestation triggering bullous pemphigoid should be distinguished from bullous pemphigoid-like scabies[14]. However it is still not clear about the mechanism responsible for the eruption and exacerbation of BP in the cases mentioned above.

To our knowledge, this is the first reported case of BP by the artificial hip made of titanium alloy. The reasons for the onset of bullous pemphigoid triggered by titanium alloy hip remain unknown but may be related to cytotoxicity of some titanium alloys containing molybdenum, niobium, and silicon [15]. Artificial hip should be added to the list of possible triggers for BP and the diagnosis should be considered if blistering develops following hip replacement surgery.

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