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Priapism as a new symptom of anaphylaxis in a prepubescent child

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To the editor,

Anaphylaxis is a severe, potentially life-threatening systemic hypersensitivity reaction. It is rapid in onset (minutes to a few hours) and typically involves multiple body systems^{1,2}. The prevalence in children is unknown, although there has been a prevalence of between 0.05 to 2% in the general population^{3,4}. There are several mechanisms involved: IgE-dependent immune-mediated, non-IgE-dependent immune-mediated, non-immune and idiopathic. Clinical symptoms of anaphylaxis are caused by release of histamine, leukotrienes, tryptase, prostaglandins, platelet activating factor and other mediators of systemic circulation (mast cells and basophils), which act on different target organs and are responsible for the clinical manifestations^{2,5,6}.

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On the other hand, priapism is a penile erection unrelated to sexual stimulation or that persists after its conclusion. It is produced by two mechanisms: Ischaemic (veno-occlusive) or non-ischaemic (arterial). The ischaemic form is the most common and manifests as a painful erection of maximum rigidity with no cavernous blood flow. If it lasts more than four hours it is considered a compartment syndrome and requires urgent medical intervention⁷. The arterial form produces no pain and is caused by uncontrolled entry intracavernous blood flow. It is rare in children and so far has been reported in relation to severe diseases such as sickle cell disease, leukaemia, diabetes mellitus I, neurological disorders or infections. Its main treatment is alpha-adrenergic agonists such as phenylephrine (the treatment of choice because it is a pure alpha-1 adrenergic agonist), adrenaline, noradrenaline and/or ephedrine⁷⁻⁹.

We report a case of a patient with priapism secondary to an anaphylactic reaction following administration of house dust mite immunotherapy. Case: Six-year-old boy diagnosed with allergic moderate persistent rhinoconjunctivitis to dust mites (total IgE 447 UI/mL; *Dermatophagoides pteronyssinus* specific IgE 70 KU/L) without other relevant medical history. First dose (0,2 mL) of a polymerized *Dermatophagoides pteronyssinus* extract was administered subcutaneously. Twenty minutes after injection, the patient developed bronchospasm (Wood Downes score 5/Moderate), rhinitis, bilateral palpebral angioedema, generalized rash, vomiting of food and associated priapism. The following drugs were administered due to symptoms compatible with anaphylaxis: intramuscular adrenaline (0.01 mg/kg), salbutamol in 3 sets of 4 inhalations each one, oral corticosteroids 1 mg/kg/dose and hydroxyzine 0.5 mg/kg/dose. After this treatment all symptoms described improved in about 25 minutes and 12 hours later the patient remained completely asymptomatic. The most frequent clinical manifestations in children with anaphylaxis include skin (erythema, urticaria and/or angioedema, the most common and first to appear), breathing (bronchospasm, dyspnoea, stridor, hoarseness, nasal congestion, runny nose, sneezing, among others), and

less frequently, cardiovascular (tachycardia, hypotension), gastrointestinal (vomiting, abdominal pain), neurological (especially sick) and/or eye-related. Other symptoms such as metrorrhagia and uterine contractions have also been described in women and prepubertal girls, although their occurrence is rare^{1,2,6}. So far, no involvement of male sexual organs has been described in children or adults.

Based on an exhaustive review of the literature, up to now this is the first case of priapism associated with anaphylaxis. The priapism originated simultaneously with the rest of clinical symptoms compatible with anaphylaxis and resolved after administration of salbutamol and adrenaline, both of which are indicated for the treatment of anaphylaxis but also reported as effective for treating priapism⁷⁻⁹. Adrenaline acts as an agonist of alpha receptors and adrenergic beta-1 and beta-2 receptors while salbutamol acts only against beta-2-adrenergic agonists. The time-course of events suggests that priapism was part of the anaphylactic reaction, rather than an adverse reaction caused by the treatment for anaphylaxis. The heterogeneous manifestations of anaphylactic reactions may hamper its recognition by clinicians; therefore, it is of importance to report unusual signs such in this case of a paediatric patient with prepubertal priapism in the context of anaphylaxis.

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