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Histaminic Angioedema in Children: A Rare Entity Not to be ignored

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Abstract **Case Report**

Angioedema is a common symptom to different etiologies, the diagnostic orientation is based on the associated signs. The most common are histamine, non IgE-mediated, benign and often associated with urticaria. Angioedema by anaphylaxis and bradykinin angioedemas are rare but potentially severe by upper airway involvement. We report the case of a 14 years old patient with a history of bilateral orbital swelling associated with deep urticaria, and a brother followed for angioedema. Presenting bilateral palpebral and jugal edema, associated with uticarian tasks in the trunk and back. The diagnosis of histaminic angioedema was made and confirmed by the response to corticosteroids and antihistamines, and a biologic workup ruling out differential diagnoses. Treatment was based on the management of the crisis and the implementation of in-depth treatment in the event of recurrent and disabling crises. Measures associated with this drug treatment are also essential, mainly therapeutic education.

Keywords: Angioedema-histamine-bradykinin-child-anti-histamine.

INTRODUCTION

Angioedema is a common condition characterised by localised and transient swelling of the subcutaneous or submucosal tissues. Angioedema may be caused by three main mechanisms: histaminic, bradykinic and idiopathic. In this study, we report the observation of a 14-year-old girl with histaminic angioedema, after free and informed consent from her parents.

OBSERVATION

Daughter CC, 14 years of age, from a non-consanguineous marriage, with a history of hospitalisation for bilateral orbital swelling associated with urticarial lesions, treated as stage I orbital cellulitis; her brother was treated for angioedema and put on antihistamines, resolved at the age of 18.

She presented with sudden onset bilateral palpebral and jugal oedema, associated with erythematous, non-itchy, non-confluent, non-inflammatory patches on the trunk and back (Figure 1), with no respiratory involvement. Questioning excluded the notion of exposure to a known allergen or medication. In addition, the patient had been suffering from urinary burning for 3 days.

Biological tests showed a CRP of 28 mg/l, without hyperleukocytosis or hypereosinophilia. Urine cytobacteriology showed hyperleukocyturia. She was started on hydrocortisone hemisuccinate 5 mg/kg/6h, combined to cetirizine 10 mg/d, and Ceftriaxone 50mg/kg/d for the urinary tract infection. Progression was marked by complete disappearance of the oedema and urticarial lesions without scarring 12 hours after the start of treatment (Figure 2).

Given these symptoms, we suggested an allergic reaction, hereditary bradykin angioedema, histamine angioedema, and a systemic disease such as vasculitis or lupus.

A biological work-up showed a normal C1 inhibitor level of 0.35 g/L, a normal C3 level of 1.25 g/L and a normal C4 level of 0.279 g/L, and an immunological work-up including: Anti SSB/ SSA/ native Anti DNA/ Antiphospholipid IgM and IgG/ rheumatoid Fc, were negative.

The diagnosis of histaminic angioedema, probably decompensated by the urinary tract infection, was made in view of: the response to corticosteroids and antihistamines, and the etiological work-up excluding other diagnosis. The patient was started on Cetirizine 10

mg/d for 3 months, combined with corticosteroid therapy at a dose of 1 mg/kg/d for 15 days with gradual degression, combined with adjuvant treatment. Progression after 3 months was marked by total disappearance of symptoms without relapse.

Figure 1: (A) Palpebral and jugal oedema, (B) Uticarial lesions in the trunk.

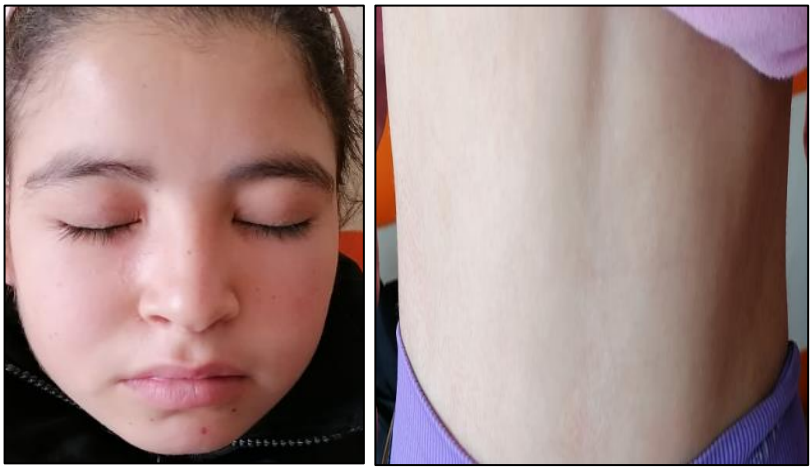

Figure 2: Clearance of oedema and lesions after 12 hours of treatment

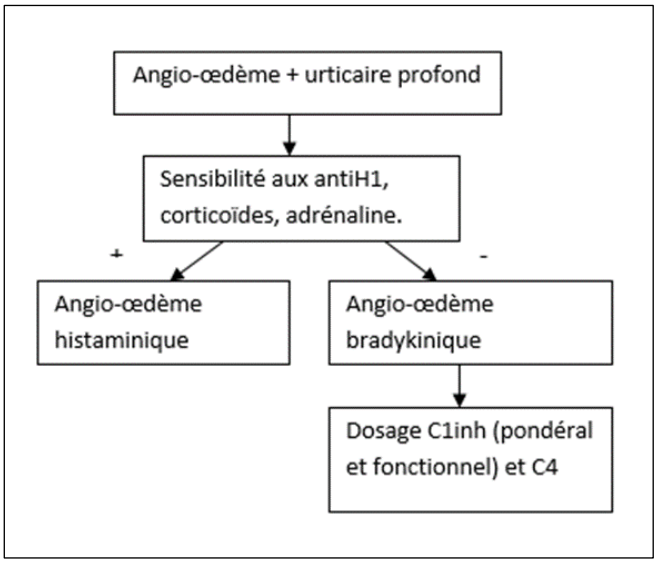

Figure 3: Algorithme for angioedema

DISCUSSION

The exact incidence and prevalence rate of paediatric angioedema is not well known, and any reported rate will clearly vary according to whether angioedema is considered with or without urticaria, or according to a specific cause of angioedema. Two studies conducted in Japan and Denmark indicated that the prevalence of non-hereditary angioedema was 4.9 and 7.4% respectively. Other studies have indicated that 15 to 25% of the population will suffer from urticaria/angioedema at some point in their lives [1]. It is difficult to judge the exact prevalence of histamine-induced angioedema. It has been estimated that the lifetime prevalence of all types of urticaria is between 8.8% and 10.8% [2]. The main cause of angioedema is histamine, due to IgE-dependent (allergy) or non-IgE-dependent mast cell activation [3, 4].

Histamine angioedema is similar to urticaria: it is the clinical manifestation of deep urticaria affecting the hypodermis or deep dermis. The colour of the skin is normal or discreetly pink, and it is not pruritic. It may be associated with a sensation of subcutaneous tension, or even pain or burning. It disappears without sequel but may reoccur. The onset is sudden and brief, generally lasting less than 24 hours. Some episodes have been described as lasting up to 72 hours. This is consistent with our study, in which the oedema and urticaria completely regressed 12 hours after the start of treatment, with no sequelae or scarring.

When a patient presents with angioedema in the emergency department, there is no rapid diagnostic test to determine the histamine or bradykin mechanism. The clinical features described above should be used as a guide. In our case, the patient had a brother who had been treated for angioedema, which may be in favour of hereditary bradykinism, but the response to corticosteroids and antihistamines pointed to histamine. The remainder of the work-up should include tests to rule out other differential diagnoses, and to find the triggering factor. These include determination of the C4 fraction of complement, determination of the weight and function of C1 Inh during the attack, and determination of C1q [5]. (Figure 3). In our case, all the tests were normal, which is in favour of the histaminic origine.

Treatment is based on conditioning, checking that the upper airways are clear, bolus of methylprednisolone 15 to 30 mg/kg (or 1 mg/m^2), in the event of respiratory distress, adrenaline is indicated in emergency treatment, in hospital, it is used at a dose of 0.01 mg/kg intravenously or intramuscularly. Basic treatment is based on antihistamines for 3 months, preferably second-generation antihistamines, which have fewer sedative and anticholinergic effects. The molecules used are Loratadine or Cetirizine, at a dose of 10 mg/d from 2 years and over 30 kg, or Cetirizine at a dose of 2.5 mg twice a day from 2 to 6 years and 5 mg twice a day from 6 to 12 years [6]. In our case, we started the patient on cetirizine 10 mg/d, because it was affordable in our context, in combination with corticosteroids 1 mg/kg/d, and an adjuvant treatment, for 15 days with progressive degression over a total period of 4 to 6 weeks.

CONCLUSION

Histaminic angioedema in children is a rare condition, which should be recognised in the presence of acute deep urticaria responding to corticosteroids and antihistamines. In-depth treatment combined with parental education is essential for long-term management.

Conflict of Interest: None

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