An uncommon Steven-Johnson syndrome induced by aBeta-Agonist agent in a 6-years-old child

*M.C. Lenti[1,2], *N. Lombardi[1,2], B. Faginali[3],V. Maggini[1,2], A. Pugi[1,2], M. Moschini[1,2], R. Bonaiuti[1,2], E. Gallo[1,2], A. Mugelli[1,2], A. Vannacci[1,2]

[1] Dept. of Neurosciences, Psychology, Drug Research and Child Health (NEUROFARBA), University of Florence
[2] Tuscan Regional Centre of Pharmacovigilance of Florence
[3] Community Pharmacy, Florence, Italy
**This two authors contributed equally to the work*

Introduction

Stevens-Johnson Syndrome (SJS) is a severe idiosyncraticimmune-mediated reaction characterized by extensive cutaneous and mucosal epidermal necrosis and sloughing. Drugs most frequently involved in SJS are antimicrobials, NSAIDs and antiseizure drugs, through a dose-independent mechanism triggered by some kind of immune response. SJS is characterized by mortality rates between 1% and 30%, depending on the extent of blistering, and up to 35% of serious sequelae in survivors. The precise incidence of SJS in children is still unknown.

Case description

A 6-year-old male child was admitted to the Emergency Department (ED) with fever, dyspnea, bilateral purulent conjunctivitis, oral stomatitis, cheilitis, skin and genital blisters. For respiratory tract infection he had been taking amoxicillin/clavulanate and betamethasone 0,5mg orally and nebulized fluticasone 50mcg; for dyspnea he had been taking nebulized salbutamol 100mcg (Ventolin® HFA), 2 sprays 3 times a day for four days. A chest x-ray revealed bronchitis. Therapy given in the first hours of arrival in ED included ophthalmic tobramycin and nebulized salbutamol (4 sprays x 4). Despite these measures his clinical condition worsened, and he was transferred to the Intensive Care Unit (ICU) where Stevens-Johnson Syndrome (SJS) was diagnosed. During hospitalization the patient developed a thrombophlebitis, which was treated for 90 days with low-molecular-weight heparin. Nine months after discharge from the hospital, the patient was hospitalized for a similar event following the re-assumption of inhaled salbutamol for dyspnea and cough. A day before ED admission patient was vaccinated against tetanus, diphtheria, and pertussis. The dermatological consultation confirmed a recurrence of SJS.

Discussion

In a 2009 FDA 'Postmarketing Adverse Event Review' two cases of Stevens-Johnson Syndrome were reported after salbutamol assumption, one of these referred to a 10-years-old male child. To our knowledge, no other similar adverse reaction was previously described in literature and databases (Micromedex®; Farmadati®). The contribution from Ventolin® HFA to the occurrence of SJS in the case we described above is strongly suggested by the temporal relationship of the event and the initiation of therapy with this drug. Positive dechallenge/rechallenge episodes are present in patient's history. The causal relationship of Ventolin® HFA in this case is confounded by concomitant use of amoxicillin, which is commonly reported in literature to be associated with SJS; moreover a role of trivalent immunization (Boostrix®) cannot be excluded. Making a final clinical evaluation and in according to the objective causality assessment performed using Naranjo Scale, our case was defined as 'probable'.

References

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