Urticaria—An Unusual Presentation of Staphylococcemia

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Urticarial skin eruptions are known to be associated with bacterial, viral and rickettsial infections(1). Amongst the bacteria, *Shigella(1), Meyingococcus(1-3),* and *Streptococcus(4)* have been implicated. Staphylococcemia presenting as urticaria in childhood has not been reported to the best of our knowledge. We report two such cases.

Case Reports

Case 1: A 10 year old boy presented with an acute episode of pruritic urticarial rash alongwith high grade intermittent fever, without chills and rigors for 3 days: There was no history of worm infestation, insect bite or exposure to any chemicals or drugs. There was no previous history of urticaria in that patient or in any other member of family. The child had received antihistaminics and steroids from a dispensary. Urticarial eruptions reappeared after initial subsidence for which he was brought to the hospital. On examination, his temperature was 38.5°C. He had generalized urticarial rash. The systemic examination was unremarkable. The patient deteriorated, developed shock and expired within few hours inspite of treatment in the form of diphenhydramine, decadron and mebendazole.

His investigations revealed, hemoglobin of 12.0 g/dl, total leucocyte count of 8000/mm^3, and differential count of 82% polymorphs, 16% lymphocytes. The blood levels of urea and sugar and serum levels of creatinine, electrolytes and immunoglobulins were normal. Urine analysis revealed mild albuminuria, 100 red blood cells and 5 polymorphs per high power field. There were occasional red cell casts. Examination of stool did not reveal any abnormality. Urine culture and throat swab cultures were sterile. However, *Staphylococcus aureus* was isolated on blood culture. A final diagnosis of Staphylococcemia with urticaria leading to septic shock was made. The parents refused autopsy.

Case 2: A 3 year old boy was admit ted with complaints of cough of 7 days duration followed 3 days later by fever, pain abdomen and vomiting. General ized urticarial rash appeared one day after the onset of fever. He received albendazole, cephalexin and antihistaminics for two days by a practitioner. There was no history of passage of worms, insect bite or exposure to chemicals or drugs. There was no history of previous epidoses of urticaria in the patient or any other family member. On examination, the average built child had a temperature of 39°C, congestion of throat and generalized urticarial rash. The systemic examination was essentially normal. He was put on cloxacillin and antihistaminics. After a few hours,

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he developed swelling over face, feet, hands and penis suggestive of angioneurotic edema. The blood pressure was maintained al) throughout. After 24 hours, he was symptomatically better; rash and edema started declining and disappeared altogether after forty-eight hours. The hemogram revealed leucocytosis (18000/mm^3) and polymorphs 80% and lymphocytes 20%. The blood levels of urea, glucose and serum levels of creatinine, sodium, potassium, glutamic oxaloacetic transaminase, glutamic pyruvic transaminase, immunoglpbulins (IgG, IgM, IgE), and complement components C_3 and C_4 were all normal. Stool and urine examination were also normal. Skiagram of chest did not reveal any abnormality. Throat swabs and urinq cultures did not grow any organisms. *Staphylococcus aureus* was grown from blood.

Discussion

Infections are often an important and frequent cause of urticaria(l). Many infectious agents, *viz.,* parasites, * viruses and bacteria have been incriminated as a cause of urticaria(l-3). In a study, infection was associated with urticaria in 43% cases(5). Streptococcus and meningococcus have been reported more frequently(l-4) but there has been only one case report associated with *Staphylococcus epidermidis(6).* Although a fairly wide range of skin lesions like impetigo, folliculitis, cellulitis, toxin mediated scalded skin syndrome, scarletina eruptions and toxic shock syndrome—have been attributed to infection with *Staphylococcus aureus(7,8)* but not a single case of urticaria associated with *Staphylococcus aureus* infection has been reported. Since blood cultures of both the patients were positive for *Staphylococcus aureus,* and there was nothing contributory in history and investigations towards another cause, it is most likely that urticaria in these patients was due to this infection.

The exact mechanism of urticaria due to infectious agents is not known. It has been postulated that these agents trigger release of histamine leukotrienes, responsible for urticaria and angioedema, from the mast cells and basophils by IgE—antigen complex; or through anaphylotoxin $C3_a$, $C4_a$ generated through activation of complement system or through the kinins, *e.g.,* bradykinin(l,7). It is conjectured that *Staphylococcus aureus* might also cause urticaria through any of these mechanisms. Urine abnormality in the first patient could have been due to small metastatic foci of infection or non suppurative renal lesions(8).

The use of steroids in the first patient had masked the severity of the disease and in the absence of antibiotics, the patient deteriorated fast, went into shock and died. This raises the question whether steroids be used in acute management of urticaria? Steroids should not be used in treatment of urticaria especially where infection is suspected to be the underlying cause. These patients should only be managed with appropriate antibiotic and antihistamine.

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Antenatal Diagnosis of Grebe Syndrome in a Twin Pregnancy by Ultrasound

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Grebe syndrome is a nonlethal distinct type of dwarfism in which a relatively normal head and trunk contrasts strikingly with a phocomelic malformation of the four extremities; the severity of the anomalies progresses distally along the limbs, so that the fingers and toes loose their phalanegeal appearance and become mere knobs(l). Polydactyly is present in about 60% of cases. This disorder is inherited as an autosomal recessive trait(2). Although a number of cases have been described postnatally, we do not know of any case diagnosed antenatally. In our prevsious report of a family with Grebe syndrome, we had mentioned about the possibility of prenatal diagnosis of Grebe syndrome by ultrasound(3).

Prenatal detection of many skeletal dysplasias has been reported earlier as in anchondroplasia, spondylo thoracic dysplasia, diastrophic dwarfism, short rib-ploydactyly syndrome type II Mejewski, Ellis-Van Creveld syndrome and many others(4).

We report probably the first case in the world, of an antenatal diagnosis of Grebe syndrome in one of the twins, the mother of whom had two earlier children with Grebe syndrome.

Case Report

A 32 years old gravida-4, para-3, was subjected for ultrasound examination at 26 weeks of gestation to rule out the possibility of Grebe syndrome, as she had two earlier children affected with Grebe syndrome. The recurrence risk in the present pregnancy was 25%, as Grebe

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