Case report

Gianotti–Crosti Syndrome- the first case report from Bahrain: A rare presentation following vaccinations

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Abstract

Gianotti–Crosti Syndrome is an idiopathic nonspecific cutaneous host response. It manifests as a benign papular rash classically limited to the extremities and the face.

We are reporting a case of an eighteen-month-old healthy boy presented with an itchy papular rash of one week duration, two days following vaccinations.

Keywords: Vaccination; Paediatrics

1. Introduction

Gianotti–Crosti Syndrome is a unique (Fastenberg and Morrell, 2007) self-limiting (Chiriac et al., 2011) papular rash typically confined to the extremities, buttocks and face. It was first described by Ferdinando Gianotti in 1955 and then by both, Gianotti and Crosti, in 1957 (Chuh, 2014). Up to date, around 300 cases were reported in the medical literature, twenty-one of which were attributed to preceding vaccinations.

We aimed to report such a rare case of an infant presented with a sudden onset of extensive progressive papular eruption after routine childhood vaccinations. We followed the case report with a literature review.

2. The case

An eighteen-month-old healthy boy presented with an abrupt onset of itchy skin-coloured pimples that started on his legs, upper limbs and the perioral area over one week duration. The rash appeared two days following his first booster of Oral Polio Vaccine, Pentavalent Vaccine [Diphtheria, Pertussis and Tetanus (DPT), Hepatitis B and Haemophilus Influenza type b] and his first dose of Hepatitis A Vaccine.

The mother denied any history of fever, contact with cases of similar symptoms and family history of atopy. The rash showed minimal response to Betamethasone cream and Cetirizine as prescribed by his family physician seen in the local health centre. A week later, the eruption extended to involve his buttocks, thighs, palms, soles and lateral sides of his abdomen. Scalp, oral cavity, and chest were spared. Mother was self-medicating her child with seawater immersion which resulted in pealing of palms of hands.

On examination, there were symmetrically distributed monomorphic skin-coloured papules on the extensor surfaces of upper and lower limbs extending to the palms,
soles and the perioral area. There were few crusted excoriations over the lower extremities, see Fig. 1. Examination of lymph nodes and abdomen was unremarkable. In the follow up, we noticed papulosquamous eruption of palms and excoriations over the gluteal region, see Fig. 2.

The temporal relationship between the recent vaccinations and this rash guided us towards the diagnosis of Gianotti–Crosti Syndrome. To rule out possible pathological aetiologies, we investigated the patient for EBV antibodies, CMV antibodies, HIV antibodies, parvovirus B19 antibodies, mycoplasma pneumonia antibodies, and viral hepatitis. Serology was negative.

Management wise, we reassured the mother, and started the patient on Crotamiton cream 10% twice daily for two weeks and Chlorpheniramine 0.3 milligram kilogram~1 along with a moisturizing lotion. The rash showed spontaneous resolution in six weeks duration.

3. Discussion

Gianotti–Crosti Syndrome peaks in children below the age of four years; yet, it can affect children aged from three months to fifteen years (Chiriac et al., 2011) with an equal distribution across males and females (Chuh, 2014). Though it is rare, some cases reported adulthood presentation (Wu, 2009; Manoharan et al., 2005; Cambiaghi et al., 1995), with female predomination (Chuh, 2014).

Gianotti–Crosti Syndrome was initially linked to hepatitis B infection subtype AYW (Erkek et al., 2001). However, negative hepatitis B serology in multiple cases diagnosed with Gianotti–Crosti Syndrome suggests other possible immunological triggers. Epstein–Barr virus is the most common associated infection worldwide. Other infectious triggers can be viral: hepatitis A virus, cytomegalovirus, coxsackievirus, respiratory syncytial virus, parainfluenza virus, rotavirus, mumps, parvovirus and molluscum contagiosum; or bacterial: Bartonella henselae, Mycoplasma pneumoniae, and group A streptococci. Where a specific infection was absent, some reports documented other associations like atopy and vaccination (Wu, 2009), see Table 1. Our patient received multiple vaccines at a time; this makes it difficult to attribute such a presentation to a single vaccination. Given the fact that all other received vaccinations were actually boosters with a negative history of associated reactions; hepatitis A vaccine might be responsible for this rash. This is consistent with Sigmon et al. (2012), Kolivras and André (2008) and Metelitsa and Fiorillo (2011). Yet, type IV hypersensitivity reaction cannot be excluded, given the 48-h timeframe of rash development.

Although rash appears as a group of small, often non-pruritic, flat-topped papules or papulovesicles of sudden onset in a previously healthy child; pruritus was reported in certain cases. Papules are typically flesh or skin-coloured, coppery red, or pale pink with a tendency of being darker on legs or in severe cases. On sites of trauma, papules appear coalesced with mild scales. Hands, wrists and buttocks are the most common sites of eruption followed by limbs and face. Trunk involvement is rare; yet it is reported (Chuh, 2003). Oral mucosa, palms and soles are typically spared (Wu, 2009) even though; our patient presented with eruption involving rare areas: palms, soles and parts of the abdomen. This rash is self-limiting with an expected spontaneous recovery in six weeks (Magyarlaki et al., 1991); however, late remission may occur in 6–12 months (Chuh, 2014). Gianotti–Crosti Syndrome has a non-relapsing course; though, recurrence was documented in one case (Metelitsa and Fiorillo, 2011). Eruption can be accompanied by fever,
lymphadenopathy, hepatomegaly and splenomegaly (Wu, 2009).

Histopathologically, findings are typically unspecific: spongiosis, hyperkeratosis, subepidermal infiltration of lymphocytes, monocytes and histocytes and deep dermal perivascular inflammation (Magyarlaki et al., 1991).

Gianotti–Crosti Syndrome is self-limiting. Supportive measures are taken in symptomatic presentations. For pruritus, topical medications like emollients and calamine lotion are sufficient. Sedative antihistamines can be added if needed. Parental education is important; aetiology and prognosis should be discussed. In cases associated with viral infections; especially hepatitis B virus, monitoring is essential (Chuh, 2014).

What makes this case unique is firstly the pruritic nature and the unusual distribution of the eruption with palms, soles and trunk involvement- which, to the best of our knowledge, was never reported; secondly the development of this syndrome post-vaccinations; and thirdly having this case reported from Bahrain as first documentation in the medical literature.

4. Conclusion

Gianotti–Crosti Syndrome following vaccine is rare. We reported a case of Gianotti–Crosti Syndrome with unusual symptomatic distribution of rash following vaccinations. Spontaneous remission occurred within six weeks.

Conflict of interest

None declared.

Table 1
Reported cases of Gianotti–Crosti Syndrome in association with vaccination.

<table>
<thead>
<tr>
<th>Report</th>
<th>Clinical presentation</th>
<th>Age</th>
<th>Vaccination</th>
<th>Interval in days</th>
</tr>
</thead>
<tbody>
<tr>
<td>Retrouvey et al. (2012)</td>
<td>Asymmetrically distributed pink papules which coalesced into plaques+ post vaccination fever</td>
<td>19 months</td>
<td>DTaP and varicella zoster virus live</td>
<td>2</td>
</tr>
<tr>
<td>Signon et al. (2012)</td>
<td>–</td>
<td>–</td>
<td>Hepatitis A and influenza</td>
<td>–</td>
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<tr>
<td>Kwon et al. (2011)</td>
<td>–</td>
<td>–</td>
<td>H1N1</td>
<td>–</td>
</tr>
<tr>
<td>Lam (2011)</td>
<td>–</td>
<td>–</td>
<td>H1N1</td>
<td>–</td>
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<tr>
<td>Kroeskop et al. (2011)</td>
<td>Symmetrically distributed flat-topped flesh-coloured papules and papulovesicles</td>
<td>9 years</td>
<td>H1N1</td>
<td>4</td>
</tr>
<tr>
<td>Atanasovski et al. (2011)</td>
<td>Monomorphic papules</td>
<td>15 months</td>
<td>MMR and DTaP</td>
<td>2</td>
</tr>
<tr>
<td>Kolivras and André (2008)</td>
<td>Symmetrical papular eruption</td>
<td>18 months</td>
<td>Hepatitis A</td>
<td>14</td>
</tr>
<tr>
<td>Monastirli et al. (2007)</td>
<td>Flesh-coloured firm 2–4 mm papules and papulovesicles</td>
<td>3 years</td>
<td>Hepatitis A</td>
<td>3</td>
</tr>
<tr>
<td>Karakaş et al. (2007)</td>
<td>A slightly pruritic skin eruption</td>
<td>5 years</td>
<td>Hepatitis B</td>
<td>21</td>
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<tr>
<td>Schwerk et al. (2005)</td>
<td>–</td>
<td>Infants</td>
<td>Poliomyelitis, DTaPs, Hib, Hepatitis B and Strep. pneumonia</td>
<td>5</td>
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<tr>
<td>Kang and Oh (2003)</td>
<td>Rash</td>
<td>3 years</td>
<td>Japanese B Encephalitis</td>
<td>1</td>
</tr>
<tr>
<td>Andiran et al. (2002)</td>
<td>Rash</td>
<td>11 months</td>
<td>Measles and the third dose of Hepatitis B</td>
<td>14</td>
</tr>
<tr>
<td>Haug et al. (2002)</td>
<td>–</td>
<td>Four infants</td>
<td>Polio Vaccine and Varicella Infection</td>
<td>4–6</td>
</tr>
<tr>
<td>Erkek et al. (2001)</td>
<td>Pupules and lymphadenopathy</td>
<td>6 months</td>
<td>DPT, Oral Polio, Hib</td>
<td>24</td>
</tr>
<tr>
<td>Murphy and Buckley (2000)</td>
<td>Flesh-coloured papules in acral distribution</td>
<td>15 months</td>
<td>MMR</td>
<td>3</td>
</tr>
<tr>
<td>Velangi and Tidman (1998)</td>
<td>Non-pruritic papules</td>
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<td>Lacour and Harms (1995)</td>
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<td>Cambiaghi et al. (1995)</td>
<td>–</td>
<td>Adult</td>
<td>Influenza</td>
<td>–</td>
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approval

The Research Committee at King Hamad University Hospital has ethically approved it.

References


