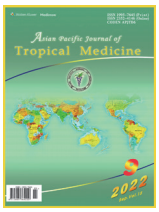

Case Report

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Acute motor axonal neuropathy following anti-rabies human diploid cell vaccine: A rare case and review

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ABSTRACT

Rationale: Guillain Barre syndrome (GBS) is an acute neurological illness leading to quadriparesis with respiratory involvement. It can be triggered by infections, vaccinations, surgery, trauma, transplantation and drugs. Anti-rabies cell culture vaccines introduced to overcome the high rate of neurological complications associated with tissue based rabies vaccine, can be very rarely associated with GBS.

Patient concerns: A 50-year-old female presented with acute severe upper back pain evolving into pure motor quadriparesis following administration of human diploid cell vaccine for rabies.

Diagnosis: Acute motor axonal neuropathy variant of GBS following anti-rabies human diploid cell vaccine.

Interventions: Intravenous high dose steroids.

Outcomes: Patient recovered completely within 1 month.

Lessons: Although anti-rabies cell culture vaccines are highly immunogenic and safe, they are rarely associated with GBS. Clinicians should be aware of this link because prompt diagnosis and treatment can result in complete recovery and avoid complications.

KEYWORDS: Immunization; Rabies; Guillain Barre syndrome; Human diploid cell vaccine; Acute motor axonal neuropathy

1. Introduction

Guillain Barre syndrome (GBS) is an immune mediated disorders leading to rapidly progressive quadriparesis with respiratory involvement. It can be triggered by infections, vaccinations, surgery, trauma, transplantation and drugs. Various vaccines have been associated with the development of GBS[1]. Rabies tissue based vaccines are associated with high incidence of neurological complications including GBS. In 1963, the adaptation of rabies virus to human diploid cell culture by Wistar Institute Philadelphia paved the way for the development of highly immunogenic and safer cell culture vaccine (CCV) for rabies prevention[2]. There are anecdotal reports of neuroparalytic illness associated with CCVs. Ours is the 5th case of GBS associated with rabies human diploid cell vaccine (HDCV) .

2. Case report

A 50-year-old female presented with 3 days history of severe upper backpain without associated motor or sensory or urinary complaints. On examination, there was no focal neurological deficit at the time of presentation. Three days into the illness, the patient developed bilateral asymmetrical facial palsy (right > left) followed by distal lower limb dysesthesia which progressed to proximodistal weakness of both lower limbs along with distal upper limb weakness in the next 2 days.

The patient had received 2 intramuscular doses of HDCV rabies vaccine following an unprovoked category ‖ dog bite over the right ankle. Second dose was administered 2 days prior to the onset of symptoms. There was no history of constitutional symptoms, gastrointestinal illness, heavy metal or toxin or drug exposure.

On examination on day 6, the patient had bilateral lower motor neuron facial palsy. Rest of the cranial nerve examination was normal. Hypotonia with hyporreflexia was noted in all four limbs with medical research council (MRC) grade 3-power proximally and 3+ distally in bilateral lower limb with moderately reduced hand grip bilaterally. Sensory examination was normal. Investigation

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revealed pure motor axonal neuropathy with normal brain and spine imaging.

Routine hematological and biochemical profile was normal. Serum C-reactive Protein, erythrocyte sedimentation rate, rhematoid arthritis factor, anti-nuclear antibodies, anti cyclic citrullinated protein antibody, protein electrophoresis, urine for porphyrinogens, antiganglioside panel IgG and IgM were all negative.

With the close temporal relationship of the development of acute onset pure motor neuropathy with the rabies vaccination, we suspected HDCV associated GBS-acute motor axonal neuropathy variant.

The patient was started on methylprednisolone 1 g Intravenously daily for 5 days as the patient refused for plasmapheresis. She started showing recovery from the 3rd day of the therapy and became completely independent in her activities by the 5th day. The patient was seen 1 month after onset of illness and had completely recovered.

3. Discussion

GBS is an immune mediated disorder triggered by an immune response to an anticident infection or vaccination where cross reactivity occurs to shared epitopes[1]. Most common implicated infection is *Campylobacter jejuni* where it was found that antibodies to specific gangliosides (including GM1, GD1a, GD1b, GQ1b) are generated in response to structurally similar oligosaccharide expressed by the bacteria[3].

Various vaccinations have also been implicated as triggers for GBS. There is limited evidence of direct causal relationship, rather the association is largely based on temporal relationship and epidemiological evidence. The strongest association has been found with 1976 swine influenza vaccine. Other vaccine associations include rabies vaccine, oral polio vaccine and tetanus toxoid[4].

High incidence (1:220) of neurological complications have been reported with rabies brain tissue derived vaccine with incidence of GBS being 1:1 600[5]. The high neurological complications and insufficient immunogenicity with the brain tissue inactivated vaccine was circumvented with development of highly immunogenic cell culture vaccines (CCV). The incidence of neuroparalytic illness is extremely low with CCVs, *i.e.* 1:32 000[6].

Only four cases of GBS with HDCV have been reported in literature. Two cases showed spontaneous recovery whereas one case was administered plasmapheresis, methylprednisolone and cyclophosphamide, showing complete recovery after 10 weeks. The fourth case original article was inaccessible[7–10].

This is the fifth case associated with CCV. Direct causal relationship could not be determined in any of the cases. Thus, though there are anecdotal reports of GBS with even CCVs, their high immunogenicity and the ability to successfully prevent development of a fatal disease, they are still highly recommended. We must be aware of these associations and report them to increase awareness, as GBS is a treatable disorder with complete recovery.

Conflict of interest statement

The authors have no conflict of interest to declare.

Declaration of patients consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Authors' contributions

TC and JS examined and worked up the case and drafted the manuscript. JS, SD and VG conceived the idea, reviewed the literature and revised the manuscript. VG supervised the case and finalized the draft.

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