CASE REPORT

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Parvovirus B19-induced acute bilateral carpal tunnel syndrome in twin girls

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We describe 2 cases of 6-year-old twin girls presenting with acute carpal tunnel syndrome (CTS) associated with human parvovirus B19 (HPV-B19) infection, as evidenced by serological data and detection of HPV-B19 DNA in blood with use of polymerase chain reaction (PCR). To our knowledge, this is the first time that HPV-B19 infection has been suggested as the causal agent of simultaneous acute bilateral CTS in twins, thus presenting the possibility that similar immunologic responses can be observed in twins during viral infections.

Keywords: Carpal tunnel syndrome; childhood; human parvovirus B19.

Human parvovirus B19 (HPV-B19) was first identified as the cause of erythema infectiosum (fifth disease) by Anderson et al. in 1983.[1] HPV-B19 is thought to infect humans exclusively,[2] and as many as 60% of adults are seropositive for HPV-B19.[2] The number of females with HPV-B19 infection is four times higher than the number of males with this infection.[3] In addition to fifth disease and asymptomatic infection, other less common manifestations of infection include anemia and pancytopenia in immunocompromised hosts, transient aplastic crisis in patients with hemoglobinopathies, nonimmune hydrops fetalis (NIHF), chronic arthritis, myocarditis, hepatitis, multisystemic vasculitis, renal disease, and idiopathic thrombocytopenic purpura (ITP).[2–5] An increasing number of reports have described HPV-B19 infection in association with a variety of neurologic manifestations.[2]

Here we describe 2 cases of twin girls presenting with symptoms of carpal tunnel syndrome (CTS) associated with HPV-B19 infection.

Case reports

Case 1 – A previously healthy 6-year-old girl presented with sudden onset of swelling of the distal extremities associated with painful paresthesia of the first 3 digits of both hands. Ten days earlier, her parents had noticed an erythematous rash, which spread from the face to the arms and abdomen, though not reaching the legs. There was no history of wrist trauma, recent immunizations, or drug use. Physical examination revealed mild swelling of the metacarpophalangeal (MCP) and proximal interphalangeal (PIP) joints. She had mild hypoesthesia in the sensory dermatomes of both median nerves, a positive Tinel's sign on the right, and a mild decrease

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in thumb abduction bilaterally. Muscle power was preserved. The acute symptoms gradually subsided after 1 week of 10 mg/kg/day naproxen sodium and high doses of vitamins B1, B6, and B12 combinations.

Case 2 – One week after the first case, her twin presented with sudden onset of swelling of the distal extremities associated with painful paresthesia of the last 3 digits of both hands. A similar history of erythematous rash was reported by the parents. Physical examination showed bilateral hypoesthesia in the sensory regions of both the median and ulnar nerves associated with swelling of the hands. It was difficult and painful to close her hands and more so to open them. Muscle power was preserved. She received the same treatments as her twin. The symptoms rapidly decreased within a few days, but numbness of the third and fourth digits persisted for 15 days.

In the first case, nerve conduction studies revealed moderate delay in distal sensory-motor latencies of both median nerves with delayed median nerve F responses on both sides. In the second case, sensory-motor conduction velocity at the wrist of both median nerves was slow, and F responses of both median and ulnar nerves were delayed bilaterally. Screening blood tests including chemistry panel, complete blood count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), rheumatoid factor, serum complement 3 and 4, thyroid-stimulating hormone (TSH), and antinuclear antibodies were normal in both cases. Rubella virus, Borrelia species, Mycobacterium tuberculosis, and other Mycobacterium species were ruled out clinically or with use of laboratory tests. Serological tests showed positive HPV-B19-specific immunoglobulin M (IgM) in both cases, and HPV-B19 DNA was detected in both patients' serum using polymerase chain reaction (PCR).

Discussion

Carpal tunnel syndrome is one of the most common peripheral compression neuropathies, but it is rarely seen in children.[6,7] Childhood CTS often has an unusual presentation, with modest complaints, and children are often too young to communicate their problem.[6] Many aspects of its etiology are not at all clear, and CTS is often termed as idiopathic; however, it has also been attributed to a variety of underlying disorders and processes.[7,8] On the other hand, CTS has been reported as secondary to infectious diseases of bacterial, mycotic, and viral origin, including HPV-B19 in adult patients.[3,8–11]

In our cases, a prior exanthematous disease associated with HPV-B19 infection is considered to be the cause of the acute bilateral CTS. Interestingly, while the symptoms of the first twin were located in the median distribution, the symptoms of the second one presented in both the median and ulnar digits. The distribution of paresthesia and pain associated with CTS is extremely variable. Symptoms are considered to be located primarily in the median distribution; however, studies have revealed that many CTS patients experience symptoms in both the median and ulnar digits more frequently than the median digits alone, as in our second patient.[12] The neurological symptoms reported in our paper are most likely due to mechanical entrapment of the median and ulnar nerves in relation to the acute arthritis associated with HPV-B19. The rapid decrease in our patients' symptoms—attributable to either non-steroid anti-inflammatory drugs or high doses of vitamin B1, B6, and B12 combinations—supports this hypothesis.

To our knowledge, this is the first time that HPV-B19 infection has been suggested as the causal agent of simultaneous acute bilateral CTS in twins, thus presenting the possibility that similar immunologic responses can be observed in twins during viral infections.

Conflics of Interest: No conflicts declared.

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