

Vaccine-derived poliomyelitis and postpolio syndrome: an Italian Cutter Incident

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Lesson

Post-polio syndrome is a condition characterized by increased muscle weakness, atrophy, fatigue and pain developing several years after the acute polio event. We describe a 52-year-old patient who experienced post-polio syndrome; he had contracted acute paralytic poliomyelitis at 12 months of age, shortly after the third dose of Salk polio vaccine.

Case presentation

A 52-year-old man was referred to our institution for muscle weakness, joint pain, progressive fatigue and ingravescent dyspnoea. These symptoms had persisted for almost 1 year and the patient was confined to a wheelchair. We observed thoracic scoliosis, waist obesity and bilateral lower limb amyotrophy, as well as electromyographic evidence of chronic partial denervation. Brain magnetic resonance imaging showed multiple hyperintense lesions in the cortical region, cortico–subcortical junction and bilaterally in the semioval centre. Blood tests, including lymphocyte subpopulations and immunoglobulin concentrations, were in the normal range. Antinuclear antibodies, antismooth muscular antibodies and rheumatoid factor were undetectable.

His clinical history showed an acute episode of poliomyelitis. At 5 months of age, he had received the first dose of Salk polio vaccine. The second dose, administered one month later, caused fever, vomiting, diarrhoea and shock. In July 1960, at 12 months of age and a few days after the third dose, the patient showed severe diarrhoeal discharges, neck stiffness and flaccid paralysis of his left lower limb. He was diagnosed with acute poliomyelitis. In June 1961, he experienced paralysis of his right lower limb. He underwent frequent hospitalization and therapeutic rehabilitation treatments.

Discussion

The interval between the administration of vaccine and the onset of clinical manifestations indicates that the Salk vaccine used to vaccinate our patient may have contained virulent poliovirus. Interestingly, our patient developed postvaccine acute poliomyelitis during a time frame (1960) that is closely connected and almost coincident to an event that occurred in the United States, known as the Cutter Incident. In April 1955, more than 200,000 children in five Western and mid-Western USA states received a polio vaccine, manufactured by the California-based family firm of Cutter Laboratories.

Within days there were reports of paralysis and subsequent investigation revealed that the vaccine had caused 40,000 cases of polio, leaving 200 children with varying degrees of paralysis and killing 10. Scientists discovered that several batches made in Cutter Laboratories contained active poliovirus. Subsequent investigation showed that the vaccine formulation contained cellular debris, which hindered adequate exposure to viral particles and did not permit complete viral inactivation by formaldehyde. This incident is regarded as one of the worst pharmaceutical disasters in the United States. The Cutter case was later revisited by several scientific journals.^{1,2} Moreover, studies have found that some batches of the polio vaccine may have been contaminated by other neurotropic viruses, including SV40 virus.^{3,4}

The new-onset symptoms of fatigue, muscular atrophy, dyspnoea, walking impairment and electromyography fulfilled, in our patient, the clinical and electromyographic criteria of post-polio syndrome (Table 1).

Post-polio syndrome is the term used for the new late manifestations that occur in patients 15–40 years after the occurrence of acute poliomyelitis. Neurological manifestations include new weakness, muscle atrophy, dysphagia, dysphonia and

Table 1. Teaching points.

1. Post-polio syndrome is a condition affecting individuals who have survived a first attack of polio. About 15–40 years after acute infection, these patients begin to present with a gradual and progressive weakness of the muscles (including those muscles not directly involved in the acute episode) as well as fatigue, muscle atrophy and joint degeneration. Skeletal abnormalities such as scoliosis may precede muscle atrophy. New muscle weakness is the most significant neurological problem.
2. This condition is thought to affect 25–50% of patients who have survived the first attack of acute poliovirus.
3. The criteria for the diagnosis of post-polio syndrome include:
(a) a previous episode of polio-induced acute flaccid paralysis,
(b) A period, generally decades, of relative clinical stability and
(c) gradual onset of new progressive muscle weakness, fatigue, muscle atrophy, muscle and joint pain, symptoms that persist for at least 1 year.
4. There is no specific therapy for post-polio syndrome. Intravenous immunoglobulin has proven effective in relieving muscle pain.

respiratory failure. Musculoskeletal pain includes muscle pain, joint pain, spinal spondylosis and scoliosis. General manifestations include fatigue, cold intolerance and sleep disorders.^{5–7} Post-polio syndrome is thought to affect 25–50% of patients who have survived the first attack of acute poliovirus.^{8,9}

The causes of post-polio syndrome have not yet been determined. Immunological studies have shown that patients with post-polio syndrome are in a chronic state of immune hyperactivation with an increase in proinflammatory cytokines. Most patients with post-polio syndrome show increased secretion of the proinflammatory cytokines IL-1 and TNF- α , which are neurotoxic and cause destruction of many nerve endings.¹⁰ However, in our patient the serum concentration of TNF- α was normal (<1 pg/mL).

There is no specific therapy for post-polio syndrome. Musculoskeletal pain is treated with weight loss, use of assistive devices, anti-inflammatory medications and physical therapy techniques. Pharmacologic agents also may be helpful, but have not been beneficial in controlled trials.¹¹ Intravenous immunoglobulin has proven effective in relieving muscle pain.¹²

Conclusion

The patient described here was affected by postvaccine paralytic polio. The actual neurologic aggravation may be included in the diagnostic criteria of post-polio syndrome (Table 1).

Declarations

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