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Prevalence and risk factors of post-polio syndrome in a cohort of polio survivors☆

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Abstract

Objective: To investigate frequency and associated factors of post polio syndrome (PPS) in an Italian cohort of people with prior poliomyelitis.
Methods: We screened subjects admitted for poliomyelitis at the paediatric hospital of the University of Palermo during the time frame 1945–1960. Patients who developed PPS were identified through a structured questionnaire and a neurologic examination. PPS diagnosis was made according to specified diagnostic criteria. Frequency of PPS was calculated in the selected cohort of polio survivors. The association with the investigated risk factors (sex, age at onset of polio, extension and severity of polio, education, associated diseases, cigarette smoking, trauma, polio vaccination) was analysed by the calculation of the odds ratio.
Results: Forty-eight participants met the adopted diagnostic criteria for PPS, giving a prevalence of 31.0%. The prevalence rate was significantly higher in women than in men ($p=0.02$). Logistic regression analyses revealed a significant inverse association with onset of poliomyelitis at over 12 months of age (OR 0.33; CI 0.14–0.79) a higher degree of education (OR 0.20; CI 0.07–0.79), and a significant association with the presence of other diseases (OR 9.86; CI 3.69–26.34).
Conclusions: In our survey one-third of patients with prior poliomyelitis had PPS. Higher age at onset of poliomyelitis is inversely associated with PPS. The association with other diseases may indicate that a chronic physical stress, particularly in already weak motor units, can contribute to the development of signs and symptoms of PPS. Our results also suggest the impact of socio-economic conditions on the risk of PPS.

Keywords: Poliomyelitis; Post-polio syndrome; Prevalence; Risk factors; Cohort study

1. Introduction

Poliomyelitis probably predates the year 1500 B.C., while the first descriptions of patients developing new neurological symptoms after acute paralytic poliomyelitis were made by Rymond and Charcot in 1875 [1]. Post-polio syndrome (PPS) became an important medical and epidemiological problem after the epidemics that occurred worldwide in the 1940s and 1950s, until vaccination became available. Although polio epidemics in the year 2003 were restricted to only six countries all over the world [2], the estimated number of persons affected by PPS is around 250,000 in Europe and 20 million worldwide because of the high number of polio survivors [3].

The pathogenesis of PPS is not completely understood. The most widely accepted hypotheses suggest a dysfunction of surviving motor neurons that causes a progressive loss of the terminals on single axons or a dysfunction of motor units, that are already weak, owing to forced exercise [4,5].

☆ This work is dedicated to Professor Giuseppe Caruso, the distinguished Italian neurologist, former Director of the Department of Neurological Sciences at the University of Naples "Federico II". Professor Caruso made important suggestions when the study project was designed. Professor Caruso died prematurely in 1999.

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Epidemiological studies investigating the prevalence of PPS are controversial, with prevalence rates ranging from 20% to nearly 100%, depending on the duration of the follow-up and the diagnostic criteria used [6–10]. Few analytical studies investigated the predictors of PPS in persons surviving acute poliomyelitis [4–6]. The present study was designed to investigate the prevalence of PPS in a cohort of patients with a previous acute poliomyelitis and to analyse whether it is associated with demographic, environmental, and clinical variables.

2. Methods

2.1. Sample

Participants in the study were identified by searching the records of the Paediatric Department and Children's Hospital of the University of Palermo. The sample frame consisted of patients discharged from the hospital with a diagnosis of acute poliomyelitis during the time period 1945–1960 ($n=575$). Each record was evaluated to confirm the diagnosis according to clinical criteria. Cases were classified according to sex, age at onset, signs and symptoms, and clinical severity based on the number of muscular segments involved. On the basis of clinical records, all patients fulfilling the adopted diagnostic criteria were included in the study. To trace each patient the resident list of the Palermo province was searched. All patients alive and resident in the study area were asked to participate. A structured questionnaire was administered and a neurological examination was performed to all subjects who agreed to participate.

2.2. Questionnaire

The questionnaire used for this study contained detailed questions about: a) the presence of muscle weakness in specific body segments; b) the presence of new neuromuscular symptoms (muscle weakness, pain, fatigue) subsequent to maximal recovery from poliomyelitis; c) changes in performing selected activity of daily living occurring within the previous ten years; d) time elapsed between acute polio and symptoms of onset attributable to PPS; e) presence of clinical disorders not related to poliomyelitis; f) cigarette smoking; g) degree of education.

2.3. Neurological evaluation and clinical criteria to define post-polio syndrome

Each patient was evaluated from both a medical and a neurological point of view. The criteria for diagnosing PPS were essentially clinical [4], namely: a) history of acute poliomyelitis; b) partial recovery of functional status followed by at least ten years of stability; c) late-onset muscle weakness with or without pain or atrophy, for which no other explanation could be found; d) exclusion of other diseases that could be responsible of the late-onset signs and symptoms. Subjects reporting only pain of radicular origin or diffuse pain without new muscle weakness or atrophy were considered to be unaffected by PPS.

2.4. Statistical analysis

We calculated the prevalence of PPS in the cohort as a percentage of the screened polio survivors. Analyses of risk factors considered the following variables: age at onset of poliomyelitis, number of affected muscles, number of affected functions (breathing, eating, swallowing, speaking, walking, running, writing, rising), smoking habits, trauma, history of other diseases, polio vaccination, and years of education. Differences between subgroup frequencies were tested by χ^2 analysis. We used the median of the distribution in the whole cohort to determine the cut-off to dichotomise continuous variables. Risk estimates between strata of a suspected risk factor were adjusted for the effect of all other factors using multiple logistic regression models. All p values were two-sided.

3. Results

Fig. 1 shows the steps we followed in our search. The entire cohort comprised 575 subjects. Eleven subjects were excluded because they did not meet the diagnostic criteria for poliomyelitis. Of the remaining 564 patients, 81 were not found into the resident list. One hundred and forty-nine subjects had emigrated and 60 were not traceable. Forty-one of the remaining 274 people had died. One hundred and fifty-five subjects of the 233 contacted agreed to be seen. Forty-eight met the adopted diagnostic criteria for PPS, giving a prevalence of 31.0%. The prevalence rate was significantly higher in women (40.3%) than in men (22.9%) ($p=0.02$; Table 1).

The mean age at onset of polio symptoms for the whole cohort was 15.4 months (median 12, range 1–84). The mean age at onset of PPS was 34.6 years (median 34, range 20–49), and the mean interval between poliomyelitis and PPS onset was 33.5 years (median 33, range 19–48.2). Muscle weakness represented the first symptom of PPS onset in 40.5% of patients, in 52.4% they started complaining muscle weakness and pain at the same time, and in 7.1% it followed pain.

Tables 1 and 2 summarize the distribution of the investigated variables among persons with previous poliomyelitis. PPS was significantly higher among patients who had the onset of poliomyelitis before 12 months of age, and it was inversely associated with a higher educational level. We also observed a significantly higher frequency of comorbid disorders in persons affected by PPS compared to polio survivors not affected by PPS.