INTERNATIONAL STUDENT CONFERENCE ON PEDIATRICS 2024

ABSTRACT BOOK

RĪGAS STRADIŅA UNIVERSITĀTE

VILNIAUS UNIVERSITETAS • 1579 • UNIVERSITAS VILNENSIS

Vilnius University

VILNIUS UNIVERSITY PRESS
2024

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Leidinio bibliografinė informacija pateikiama Lietuvos nacionalinės Martyno Mažvydo bibliotekos Nacionalinės bibliografijos duomenų banke (NBDB).

DOI: https://doi.org/10.15388/Proceedings.2024.45
ISBN 978-609-07-1049-4 (skaitmeninis PDF)

ISOPRINOSINE THERAPY IN SUBACUTE SCLEROSING PANENCEPHALITIS

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Introduction: Subacute Sclerosing Panencephalitis (SSPE) is a rare, incurable neurological disorder, caused by persistent measles infection, primarily affecting children. It is characterized by cognitive decline, seizures, and coma. Isoprinosine (IP) is the only medication with official indications for SSPE in multiple countries. Globally, other treatments, including combination therapies, are gaining popularity, therefore understanding the efficacy of IP monotherapy is crucial. This review aims to assess the effectiveness of IP monotherapy in treating SSPE, comparing outcomes based on treatment initiation stage and the time frame of case reports.

Results: We conducted a systematic search on PubMed using keywords "SSPE", "Subacute Sclerosing Panencephalitis". Inclusion criteria: SSPE treated with IP monotherapy, specified stages before treatment, outcomes. Exclusion criteria: reviews, immunocompromised patients, animal/in vitro studies, unrelated or unfinished studies. We analysed 53 cases: 35 (66%) started treatment in stage I-II, and 18 (34%) in stage III-IV. In earlier stages, progression occurred in 27 cases (77%), with only 8 (23%) cases stable or improved. Among stages III-IV, SSPE progressed in 8 cases (44%), with 10 (56%) cases stable or improved. In studies published before 1994, 43% showed stable or improved outcomes (19% in stage I-II, 24% in stage III IV), whereas in the last 30 years, no cases in our search exhibited stability or improvement.

Discussion: In our analysis, disease progression was more prevalent in st. I-II compared to st. III-IV. The outcomes varied: 43% of cases, reported before 1994, showed stabilization or improvement compared to none after 1994. Pritha et al. (2022) examined this treatment from 1999 onwards: only 5 out of 22 patients showed stabilization/improvement. This, together with our observation, suggests worsened outcomes of IP monotherapy in recent decades. We also identified a shift towards other therapies: in our initial search, focused on all types of treatment in the last 30 years, only 10 out of 34 articles described IP monotherapy. This could be due to availability of alternative treatments or challenges in conducting research on rare diseases. We excluded articles that did not specify SSPE stages – this resulted in fewer studies in our analysis. However, a review by Sliva et al. (2019) did not classify the patients according to stage or timing: out of 149 patients treated with IP monoterapy, 62 (41.6%) improved/stabilised, while 87 (58.4%) progressed. They decided that IP was clinically useful, either alone or in combination. While IP monotherapy may offer some benefit, the variability in SSPE outcomes suggests the importance of further research of other treatment strategies.

Conclusions: Starting IP monotherapy in the early stages of SSPE does not consistently result in improved outcomes. There is also a decline in reported stabilization or improvement in recent decades, compared to cases from over 30 years ago. Going forward, researchers should investigate other treatments to improve outcomes for SSPE patients, while also emphasizing the importance of measles vaccinations to prevent the development of SSPE in the first place.

Keywords: Subacute sclerosing panencephalitis, SSPE, isoprinosine, measles infection.