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Efficacy and influencing factors of most prevalent immunosuppressive therapy for pure red cell aplasia:meta-analysis and systematic review

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ADMINISTRATIVE INFORMATION

Support - None reported.

Review Stage at time of this submission - Data analysis.

Conflicts of interest - None declared.

INPLASY registration number: INPLASY202440070

Amendments - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 17 April 2024 and was last updated on 17 April 2024.

INTRODUCTION

Review question / Objective Pure red cell aplasia (PRCA) is a hematological syndrome, characterized by anemia and a marked reduction of erythroid progenitor cells with multiple etiologies. Corticosteroid (CS), Cyclosporine (CsA) and cyclophosphamide (Cyt) are the main immunosuppressive therapy (IST) used in treating acquired pure red cell aplasia (aPRCA). As PRCA is rare, the studies conducted on the above commonly used drugs also were small sample size studies. This study evaluated the efficacy and influencing factors of above agent in treating PRCA.

Condition being studied Pure red cell aplasia (PRCA) is a hematological syndrome, characterized by anemia and a marked reduction of erythroid progenitor cells with multiple etiologies. Corticosteroid (CS), Cyclosporine (CsA) and cyclophosphamide (Cyt) are the main immunosuppressive therapy (IST) used in treating

acquired pure red cell aplasia (aPRCA). As PRCA is rare, the studies conducted on the above commonly used drugs also were small sample size studies. This study evaluated the efficacy and influencing factors of above agent in treating PRCA.

METHODS

Search strategy We searched the electronic databases, including PubMed, Embase, Cochrane Library and Web of Science, up to March 31, 2024, with citations in English. The following key terms were used: Pure red cell aplasia, immunosuppressive treatment, Corticosteroid (CS), Cyclosporine (CsA) and cyclophosphamide (Cyt) and gene mutation.

Participant or population Our study population are aPRCA, including primary PRCA, secondary PRCA.

Intervention Immunosuppressive therapy.

Comparator Included literatures were cross-sectional studies or case series, so the meta-analysis was a meta-analysis of one-armed studies.

Study designs to be included PubMed, Embase, Cochrane Library and Web of Science were systematically searched. We included studies investigating the efficacy of IST in treating aPRCA. Two researchers independently screened, extracted data, and assessed the quality of the included studies using the MINORS scales. STATA/MP16 was used for meta-analysis, Overall response rates (ORR) include complete response rates (CR) and partial response rates (PR) were synthesized using either a random-effects model or a fixed-effects model and the results were analyzed for publication bias.

Eligibility criteria (1) Our study population are aPRCA, including primary PRCA, secondary PRCA, we excluded the study congenial pure red cell aplasia like diamond blackfan, we also excluded studies of aPRCA with specific etiologies, such as pregnancy-associated aPRCA, ABO incompatible stem cell transplantation-associated aPRCA, and infection-associated aPRCA, because the specific etiologies described above lead to significant differences in disease outcomes. (2) clinical trials investigating the efficacy and influencing factors of IST in patients with aPRCA. Outcomes regarding treatment responses could be extracted or calculated. (3) If the study recruited participants in the same period or in the same center, we included only the studies with the largest sample size. We excluded case reports, case series ($n \leq 5$), comments, letters, Reviews, animal research, conference abstracts and article which is not English.

Information sources PubMed, Embase, Cochrane Library and Web of Science.

Main outcome(s) Immunosuppressive therapy is effective in pure red cell aplasia.

Quality assessment / Risk of bias analysis Assessed the quality of the included studies using the MINORS scales and the results were analyzed for publication bias.

Strategy of data synthesis Overall response rates (ORR) include complete response rates (CR) and partial response rates (PR) were synthesized using either a random-effects model or a fixed-effects model.

Subgroup analysis Subgroup analysis and meta-regression were performed based on etiology, medication use (type of drug, monotherapy or in multitherapy, first-line or second-line treatment), gene mutation.

Sensitivity analysis Elimination of heterogeneous phenotypes after sensitivity analysis.

Country(ies) involved China.

Keywords Pure red cell aplasia, Immunosuppressive Treatment, Gene mutation, Meta analysis.

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