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Pallidal versus subthalamic deep-brain stimulation for meige Syndrome: a systematic review and meta-analysis

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Wu, X[1]; Xue, T[2]; Pan, SQ[3]; Xing, WK[4]; Huang, CJ[5]; Zhang, JG[6]; Zhao, GZ[7].

Corresponding author:
Guozheng Zhao

zhaoguozheng2021@suda.edu.cn

Author Affiliation:
Suzhou Ninth People's Hospital.

ADMINISTRATIVE INFORMATION

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Amendments - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 20 June 2023 and was last updated on 20 June 2023.

INTRODUCTION

Review question / Objective The efficacy and safety of deep brain stimulation (DBS) targeting the globus pallidus internus(GPi) and subthalamic nucleus(STN) for Meige syndrome (MS) have not been compared in large-scale studies. This meta-analysis aimed to identify the superior DBS target for MS treatment.

Rationale MEDLINE, EMBASE, the Cochrane Library and ClinicalTrials.gov were systematically searched to identify relevant studies regarding DBS for MS published before December 31, 2022. The primary outcome was improvement in Burke-Fahn-Marsden Dystonia Rating Scale movement (BFMDRS-M) and disability (BFMDRS-D) scores. Secondary outcomes, including baseline patient characteristics and stimulation-related complications (SRCR) after GPi-DBS and STN-DBS for MS, were also analyzed. Pearson's correlation coefficients and stepwise multivariate regression analysis were used to identify potential prognostic factors.

Condition being studied Meige syndrome (MS) is a segmental myodystonia symptomized by a combination of blepharospasm (BSP), oromandibular myodystonia (OMD) and neck muscle movement disorder. Drug treatment is a first-line treatment. However, the reaction of most patients is usually unsatisfactory, and some of them have experienced troublesome adverse reactions.We consider to treat MS with deep brain stimulation (DBS), a technique that has long been adopted for primary myodystonia. Due to its high short- and long-term efficacy, this surgery may serve as a safe and effective alternative in the treatment of MS.The globus pallidus internus(GPi) and subthalamic nucleus(STN), as two targets, can be stimulated in DBS. The safeties and efficacies of stimulating both targets have been reported, but their clinical outcomes rarely compared. To evaluate the efficacy and safety of GPI- and SPN-targeting DBS, we initiated this study.

METHODS

Search strategy MEDLINE, EMBASE, the Cochrane Library and ClinicalTrials.gov.

Participant or population patients ≥18 years of age diagnosed with primary Meige Syndrome, based on the standard clinical diagnostic criteria.

Intervention Patients treated with GPi-DBS and patients treated with STN-DBS.

Comparator Patients treated without GPi-DBS and patients treated without STN-DBS.

Study designs to be included Types of study to be included case control and cohort.

Eligibility criteria The inclusion criteria were as follows: (1) language restriction: available in English; (2) participants: patients ≥18 years of age diagnosed with primary Meige Syndrome, based on the standard clinical diagnostic criteria[23]; (3) intervention: GPi-DBS and STN-DBS; (4) outcomes: efficacy outcomes including objective scales Burke-Fahn-Marsden Dystonia Rating Scale-Movement (BFMDRS-M) including total scores and subitem scores, and -Disability (BFMDRS-D),and absolute improvement rate of BFMDRS((BFMDRSpre-BFMDRSpost)/BFMDRSpre*100%) ,the safety outcomes was stimulation-related complications. Included studies were not required to include all the outcomes mentioned above but at least report BFMDRS score. The exclusion criteria were as follows: (1) study type: conference articles, editorials, case reviews and reviews; (2) case report that included only one patient; (3) indications for surgery other than Meige Syndrome; (4) a stimulation target other than GPi or STN.

Information sources MEDLINE, EMBASE, the Cochrane Library and ClinicalTrials.gov were systematically searched to identify relevant studies published before December 31, 2022. Two independent investigators systematically searched the above four databases with the search strategy of ("globus pallidus interna" OR "subthalamic nucleus" OR "deep brain stimulation") AND "Meige Syndrome" in title, abstract or keywords. Additionally, the reference lists of included studies, relevant systematic reviews and meta-analyses were also screened independently and manually to ensure a more comprehensive search.

Main outcome(s) Burke-Fahn-Marsden Dystonia Rating Scale-Movement (BFMDRS-M) including total scores and subitem scores, and -Disability (BFMDRS-D),and absolute improvement rate of BFMDRS((BFMDRSpre-BFMDRSpost)/BFMDRSpre*100%.

Additional outcome(s) stimulation-related complications.

Quality assessment / Risk of bias analysis The Methodological Index for Non-randomized Studies (MINORS) was used to assess the quality of each included study. MINORS comprises 12 items dealing with potential areas of bias, with the first subscale of eight items dealing with non-comparative study while all 12 items were relevant to comparative studies. Each item receives a score from 0 to 2, resulting in the ideal global score would be 16 for the non-comparative studies and 24 for the comparative studies. The assessment was carried out separately by two investigators . Disagreements between the two investigators were resolved through consensus or by another independent investigator.

Strategy of data synthesis We used Review Manager 5.3 software to perform pairwise meta-analysis of direct evidence. The mean difference (MD) with 95% confidence intervals (95% CIs) were analyzed and calculated using a random effect model for the continuous outcomes. We then estimated heterogeneity through the I2 statistic as follows: I2 < 30% suggests "low heterogeneity"; I2 between 30% and 50% indicates "moderate heterogeneity"; I2 > 50% denotes "substantial heterogeneity".

Subgroup analysis To test whether there is any difference in the postoperative improvement of efficacy outcomes between the GPi-DBS and STN-DBS groups, GraphPad Prism 8.0 software (GraphPad Software, San Diego, CA, USA) was used for statistical analysis. All of the data are presented as the mean ± SD. Data sets in each group were tested for normality of distribution with the Kolmogorov–Smirnov test, and all data exhibited a normal distribution. Data were analyzed by the two-tailed Student's t test to compare differences between two groups. We also used two-tailed Pearson correlation analyses to determine if there was a correlation between the data of two groups. Meta-regression analyses were also performed to examine a linear association between BFMDRS-M score and the outcome of interest, using a random-effects model and illustrating, if any, regression line and its 95% prediction intervals. For all the analyses, a P value < 0.05 was considered to be statistically significant.

Sensitivity analysis Meta-regression analyses were also performed to examine a linear association between BFMDRS-M score and the outcome of interest, using a random-effects model and illustrating, if any, regression line and its 95% prediction intervals. For all the analyses, a P value < 0.05 was considered to be statistically significant.

Country(ies) involved China.

Keywords Meige syndrome.globus pallidus internus.subthalamic nucleus.meta-analysis.BFMDRS-M.BFMDRS-D.

Contributions of each author
Author 1 - Xin Wu.
Author 2 - Tao Xue.
Author 3 - Shiqing Pan.
Author 4 - Weikang Xing.
Author 5 - Chuanjun Huang.
Author 6 - Jianguo Zhang.
Author 7 - Guozheng Zhao.