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Tübingen Hip Flexion Splint for Developmental Dysplasia of the Hip in Children and its Safety Assessment: A Systematic Review and Meta-Analysis

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Abstract

Background

Hip dysplasia is an abnormality of the hip joint where the socket portion does not fully cover the ball portion, which might increase the risk of joint dislocation. Hip dysplasia may occur at birth or develop in early life. The purpose of the present study was to assess the effect of treatment with Tübingen hip flexion splint for Developmental Dysplasia of the Hip (DDH), and its safety.

Materials and Methods: The systematic search was carried out on the online databases (Medline, Cochrane library, EMBASe, and Scopus) to assess the effect of treatment with Tübingen hip flexion splint for developmental dysplasia of the hip. Articles indexed until 20 April 2020 were reviewed using keywords such as (Hip dislocation, Congenital, DDH, and Tübingen hip flexion splint). The study selection was carried out by two reviewers.

Results: Six studies were included in systematic review and meta-analysis. The pooled successful rate of Tübingen hip flexion splint was 88% with a Confidence Interval [CI]; 87-97%; p<0.001; heterogeneity; I2: 89%%, p<0.001; six trials; random effect model. The heterogeneity level was high among the included studies. Due to high heterogeneity, the sensitivity analysis was conducted to assess the effect of each study on the result and the level of heterogeneity. The results of the sensitivity analysis showed that no studies influenced the outcome and the level of heterogeneity. However, the pieces of advice were not associated with serious side effects.

Conclusion

According to the results, the success rate of this Tubingen brace was 88%. Moreover, the results must be interpreted with caution due to the small sample size and high heterogeneity. The effectiveness of this method in the long term is still debatable.

Key Words: Children, Development, Hip dysplasia, Tübingen hip flexion splint.

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1- INTRODUCTION

Developmental Dysplasia of the Hip (DDH) is one of the most common disorders affecting the neuromuscular system of children due to several unclear factors (1). Genetic factors are involved in the development of this disease so that each parent's involvement increases the risk of the children's involvement up to 10 times. Notably, 80% of infants with the Congenital Dislocation of the Hip (CDH) are girls (2). The risk of breech position among the CDH patients is about 20%, while this trend is about 2-4% among the healthy population. Oligohydramnios and neuromuscular diseases are associated with an increase in CDH risk (3). Swaddling also increases the risk of CDH (4). In general, disorders in the left hip are more common compared to the right hip (5). The CDH is often caused by joint capsule loosening (5). When the femoral head of a baby with CDH stays out of the acetabulum for a while, both the growth of the femoral head and the acetabulum are impaired. Moreover, the femoral head remains small and does not grow well. The femoral neck remains thin, and the femoral shaft will be short in diameter (6).

If the infants are born later, and the treatment onset is delayed, these changes become more difficult to be treated. Early manifestations of the disease are associated with clinical signs of Ortolani and Barlow's test. Gluteal sulcus, decreased abduction, and affected limb shortening can appear gradually. Simultaneous clinical examination and ultrasound, especially in the first months of life, help to accurately and quickly diagnose the CDH (7). These patients are treated with a variety of surgical treatments, hip reduction, and joint stability (8). Pavlik Harness (PH) is the most common and preferred non-surgical treatment among infants with DDH (9). The success rate of this method varied from 25-100%. This meta-analysis aimed to assess the effect of treatment with Tübingen hip flexion splint for DDH and its safety.

2- MATERIALS AND METHODS

2-1. Information sources

Systemic search on Medline (via PubMed), Cochrane Library, EMBASE, and Scopus were carried out and articles indexed until 20 April 2020 were reviewed using keywords such as (Developmental Dysplasia of the Hip (DDH), Hip dysplasia, Congenital Hip Displacements, Congenital Dysplasia of the Hip, Congenital hip dislocations, and Tübingen hip flexion splint). All studies which assessed the Tübingen hip flexion splint for developmental dysplasia of the hip in children were included in this study. The search was carried out independently in duplication by two reviewers, and the supervisor dissolved any disagreement between the reviews. Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) checklist was used as a template for this review.

2-2. Inclusion Criteria

Participants, Interventions, Comparators, and Outcomes (PICO) was used to formulate the review objective and inclusion criteria.

Participant: Children up to 3 years old.

Interventions*:* All studies used of advice of Tübingen Hip Flexion Splint for developmental dysplasia.

Comparators*:* All studies were performed as pre-post-surgery without control group.

Outcome: Primary outcome of our systematic review is to assess successful rate and secondary outcome included safety of Tübingen Hip Flexion Splint.

2-3. Included Studies

Randomized controlled trials (RCT), clinical studies both randomized and non-randomized, either retrospective or

prospective. Due to the limited number of published RCT in the literature, other types of clinical studies were included. Pilot, preliminary, and case report studies were not included due to limited sample size and a higher risk of bias—studies published in English until April 2020.

2-4. Quality of Study

Two authors assessed the methodological quality of the studies. Newcastle Ottawa Scale (NOS) was used to evaluate critically appraise the quality of non-randomized studies in meta-analyses. The scale has three domains: selection (0-4 points), comparability (0-2 points), and ascertainment of outcome (0-3 points) (10).

2-5. Data Extraction

Table. 1 showed the characteristics of five included in the meta-analysis. These included Study, Country, Year, and Number of hips, Study design, Successful rate, Side effect, s and Newcastle Ottawa Scale (NOS).

2-6. Statistical Analysis

All statistical analyses were performed using Comprehensive Meta-analysis Version 2.0 (Biostat, Englewood, NJ, USA). For the heterogeneity assessment, Cochrane Q-test ($p< 0.05$ as statistically significant), and the I2 index were used.

3- RESULTS

3-1. Baseline characteristics

Baseline characteristics of five studies included into systematic review showed in **Table.1**. Rapid and effective diagnosis and treatment of DDH is of particular importance in order to prevent future surgical interventions (11). Finally, the systematic review and meta-analysis were conducted to analyze six articles (12-17). Munkhuu et al., reported the treatment of 36 Type 2c (0.2%), and 1715 Type 2a (10.3%) of hips using a Tubingen hip flexion splint. The hip disorders were removed in all children who were followed up (12). In a study by Kubo et al., 95.4% of Tübingen splint-treated hips were converted into a type I hip following the mean treatment duration of about 89 days (13). Atalar et Al. reported the success rate of 93.3% in the treatment of DDH hips (14). In a study by Pavone et al., the successful transfer into type I hips was reported for 90.44% of dysplastic unstable or dislocated hips having the α-angle of more than 64° in the splint (15). In a study by Seidl et al., the successful transfer into type I hips was seen in 98% of the dysplastic unstable or dislocated hips were successfully converted into type I hips having the α-angle of more than 64° in the splint (16). In a study by Yegen et al., the cut-off point of the onset of primary therapy was the 15th week, with 62.50% specificity, and 84.62%, sensitivity, respectively. The success rate of 75% was seen in 78 hips having the satisfactory outcomes (17).

3-2. Meta-analysis

Six studies were included in systematic review and meta-analysis. The pooled successful rate of Tübingen hip flexion splint was 88% with a 95% Confidence Interval (CI); 87% to 97%; $p<0.001$; heterogeneity; I2:89%%, $p<0.001$; six trials; random effect model (**Figure. 1**). The heterogeneity level was high among included in studies. Due to high heterogeneity among studies, a sensitivity analysis was conducted to assess the effect of each study on the result and degree of heterogeneity. The results of the sensitivity analysis showed that none of the studies influenced the outcome and the level of heterogeneity (**Figure. 2**).

Table-1: General characteristic of studies included into meta-analysis, and total score of methodological quality.

Author, Year, Country, Reference	Numbers hips	Study design	Successful rate	Side effects	NOS
Munkhuu et al., 2013, Mongolia, (12)	1751	Perspective	100%	Evidence for severe treatment related complications	8
Kubo et al., 2018, Germany, (13)	109	Perspective	95.4%	Not mentioned	8
Atalar et al., 2014, Turkey, (14)	60	Perspective	93.3%	Dysplasia in early but later resolved in 4 patients.	8
Pavone et al., 2015, Italy, (15)	10274	Perspective	90.44%	Avascular necrosis	8
Seidl et al., 2012, Germany, (16)	50	Perspective	98%	Not mentioned	8
Yegen et al., 2019, Turky, (17)	104	Perspective	84.62%	Not mentioned	8

Newcastle-Ottawa Scale (NOS).

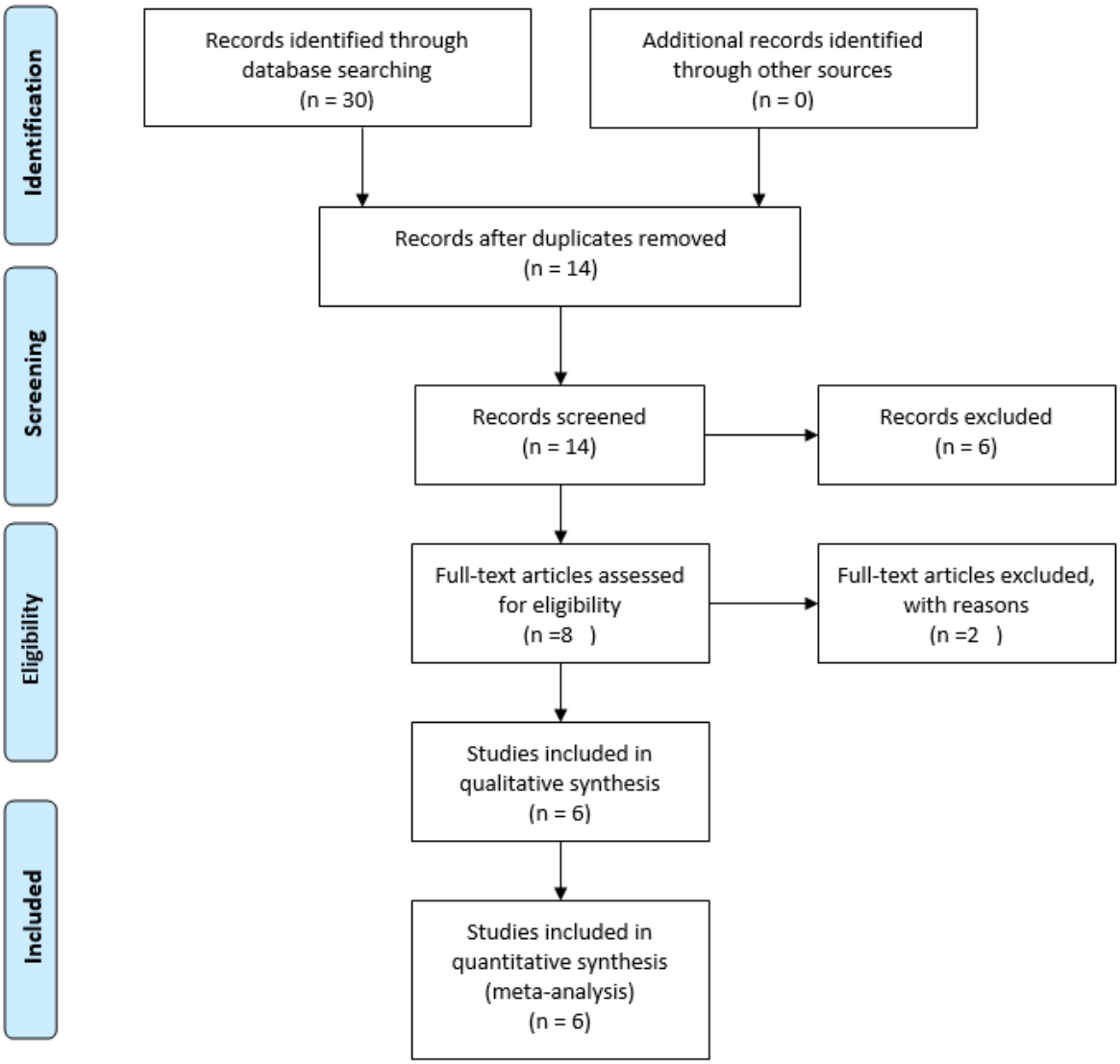

Fig.1: PRISMA flowchart.

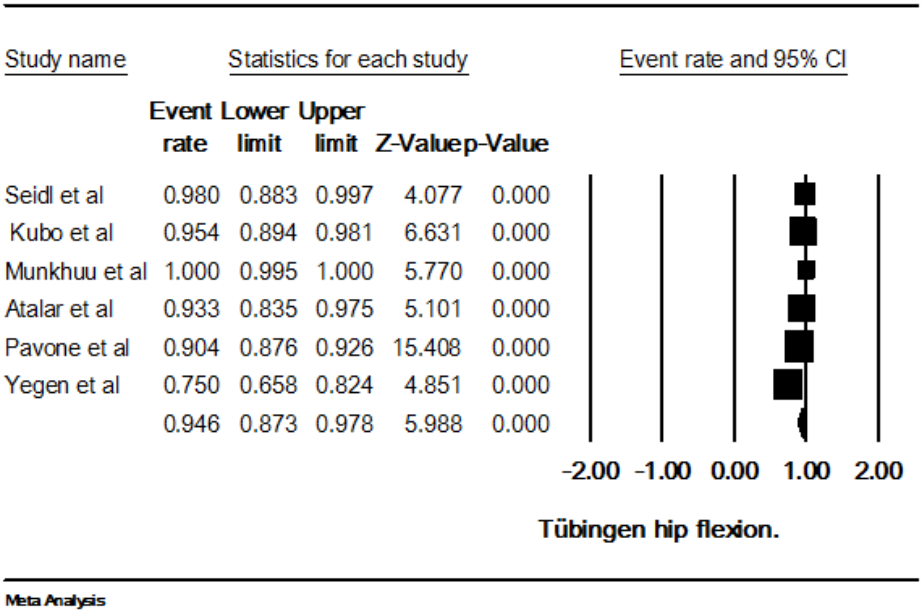

Fig.2: Tübingen hip flexion splint for developmental dysplasia of the hip in children. The midpoint of each section of the line approximates the mean and line breaks represent a 95% confidence interval in each study.

4- DISCUSSION

This meta-analysis aimed to assess the effect of treatment with Tübingen hip flexion splint for DDH and assess its safety. DDH is an exceptional condition that affects children worldwide. It has been suggested that screening programs should be prioritized in different countries to diagnose and treat the disease early. Rapid and effective diagnosis and treatment of DDH is of particular importance to prevent future surgical interventions (11). There is no consensus on the best procedure to treat this disorder. Various devices have been introduced and proposed and are still being updated along with the advances in medical science (2). Pavlik Harness (PH) is the most common and preferred non-surgical treatment in infants with DDH. If this method is successful, the anatomical position of the hip and its function will improve, especially at fewer than six months old (9). However, several parameters affect the success of the PH method, including age, sex, laterality, DDH family history, breech presentation, intrauterine packing, first-born girl, oligohydramnios, and swaddling. Therefore, the success rate of this method is not precisely determined by the influence of several factors, and the number of clinical studies with a high level of documentation is limited in this regard. The effectiveness of this method, both in the short and long term, is still debatable, given the variables associated with the patient, and needs to be carefully and comparatively studied using other methods (18). The von Rosen splint is another method used to treat DDH. The reported risk of Avascular Necrosis (AVN) results from this method varies from 0.2-1.8. However, faster initiation of treatment with this method (less than three months) may prevent the severity of the complication, while the researchers have not yet provided any evidence to support this. Therefore, due to the heterogeneity in several studies, which makes it impossible to conclude whether age at the time of treatment is the leading and determining factor in AVN risk. The safety of this tool has not been well supported in studies. Designing a controlled prospective study with regular follow-up of radiography at

skeletal maturation appears to be helpful (19). Tubingen brace is another method. According to our meta-analysis, the success rate of this method was 88%. The failure of the treatment is mainly due to improper use of the splint, which can lead to neurological injuries to the femur due to excessive bending, AVN due to extreme deviation, and unstable reduction due to weak flexion and bending, especially in families with low socioeconomic status. Parental non-compliance is one of the main reasons for the failure of this method (15). Therefore, it is recommended that a qualitative study should be conducted to determine the reasons for parental non-compliance.

4-1. Limitations of the study

One of the main limitations of this study was the moderate to high heterogeneity between studies, and sensitivity analysis failed to show the cause of the heterogeneity. Due to the small number of studies, it was impossible to assess the heterogeneity using meta-regression. Furthermore, another limitation was the impossibility of evaluating publication bias due to the small number of articles. The methodological quality was weak in some of the studies in this systematic review. Other limitations of this study include the small number of studies and their modest sample size, indicating the need for more studies with a larger sample size in this field. The results of some studies with a small sample size (16), may change if their sample size increases. Future studies should be carried out with a large enough sample size and a control group.

5- CONCLUSION

According to our meta-analysis, the success rate of the Tubingen brace method was 88%. The findings of this study should be interpreted with caution due to the small sample size and high heterogeneity. It is suggested for further clinical trials with a larger sample size and comparison between several methods to determine the suitable device from these five devices (the Pavlik harness, the Von Rosen splint, the Tubingen brace, the Frejka pillow, and the Aberdeen splint) for physicians and parents.

6- CONFLICT OF INTEREST: None.

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