Dorsal shortening vs. ventral lengthening procedure for correction of congenital ventral curvature in patients with and without severe hypospadias: A meta-analysis of comparative studies

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ABSTRACT

Introduction: Debates remain on the ideal congenital ventral curvature correction among patients with and without severe hypospadias. Herein, we aimed to assess the comparative surgical outcome of dorsal shortening (DS) vs. ventral lengthening (VL) procedures for correcting congenital ventral curvatures.
Method: A systematic literature search was performed in September 2021 using the PubMed, EMBASE, Scopus, CENTRAL, ProQuest, and Clinicaltrials.gov databases. Comparative studies were identified and evaluated according to Cochrane Collaboration recommendations. Assessed outcomes included success and complication rates, which were extrapolated for the respective odds ratios (OR) with 95% confidence intervals (CIs). Subgroup analyses were performed according to congenital curvature, with or without severe hypospadias or recurrent curvatures (International Prospective Register of Systematic Reviews (PROSPERO): CRD42021276193).
Results: Based on pooled effect estimates from 12 studies with 430 (DS 253, VL 177) cases of ventral curvature repair, VL was able to render a better success rate for curvature correction (OR 4.20, 95% CI 2.11, 8.33) than DS repair, with comparable composite surgical complication rates (OR 0.77, 95% CI 0.27, 2.18). Furthermore, subgroup analysis showed that the success rate remained significantly better for the VL approach among patients with associated severe hypospadias (OR 3.59, 95% CI 1.25, 10.26)

and recurrent penile curvatures (OR 5.70, 95% CI 1.69, 19.21), but not among those with congenital curvature without hypospadias or those with mild hypospadias (OR 2.99, 95% CI 0.32, 27.57).
Conclusions: In correcting congenital curvature associated with severe hypospadias and recurrent curvatures, VL procedures might render a modestly better success rate; however, careful selection of appropriate patients seems to be the crucial key to the best outcome. The surgical complication rate seems to be comparable between the two approaches.

INTRODUCTION

The congenital ventral curvature of the penis can be an isolated condition, although it is more commonly associated with severe hypospadias (1). Owing to the long-term functional issues associated with penile curvature, correction of the ventral curvature is considered a critical step in hypospadias repair (2). Furthermore, many complications from hypospadias repair are associated with recurrence or inadequate curvature correction (3, 4). Clinically significant penile curvature >30° has been reported to negatively affect future sexual functionality and satisfaction (2). Various surgical correction approaches have been reported in the literature, with the majority advocating for dorsal shortening (DS) because of its simplicity and minimally invasive nature compared with ventral lengthening (VL), which involves urethral mobilization and corporotomies with or without grafting/flap utilization (1-4).

Ongoing debates remain regarding the correction of clinically significant (>30°) congenital penile ventral curvature in patients with and without severe hypospadias. Herein, we aimed to compare the surgical outcomes of the DS and VL procedures for the correction of congenital ventral curvatures through a systematic review and meta-analysis of the available literature.

METHODS

The review was registered *a priori* in the PROSPERO registry (CRD42021276193) and performed according to the Cochrane Collaboration recommendations (5). In addition, the reporting of this systematic review and meta-analysis was guided by the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) Statement and domains described by A MeaSurement Tool to Assess systematic Reviews (AMSTAR-2) (6, 7).

Population, intervention, comparison, and outcome assessment (PICO)

This systematic review and meta-analysis focused on the pediatric population with congenital penile curvature with or without hypospadias that underwent surgical correction. Specifically, for primary repair of congenital curvature at least 30 degrees, staged hypospadias repair due to severe ventral curvature, or recurrent ventral curvature correction. The intervention and comparator surgical approaches were ventral lengthening (which involves urethral mobilization and corporotomies with or without grafting/ flap utilization) and dorsal shortening (including techniques of dorsal plication). The primary outcome for comparison includes the curvature improvement as success rates per intervention

groups on the respective studies' most extended follow-up data reported; an additional outcome assessed was the composite complication rates related to the intervention.

Identification of the literature

A systematic literature search was performed in September 2021 using a medical library reference service specialist. The electronic databases used for the literature search were PubMed, EMBASE, Scopus (limited to medicine), PubMed, and Cochrane Library. Furthermore, ProQuest dissertations and theses globals were searched for gray literature. No language restrictions were applied. This was performed to capture all relevant studies and minimize possible reporting and publication bias. Relevant review articles were also identified for cross-referencing to identify the eligible titles. When feasible, communication with the corresponding authors of the included studies was attempted to clarify any missing or ambiguous information. Appendix B details the platform-specific and topic-sensitive search strings used in each electronic database.

Evaluation of the literature and publication bias

Two reviewers (MEC and PY) independently reviewed the retrieved records to screen and identify citations of all comparative studies in the pediatric literature that assessed the surgical outcomes of significant congenital ventral penile curvature with or without hypospadias. Comparative studies that assess the surgical intervention of DS (plication techniques) versus VL (corporotomies with or without graft/flap utilization) were considered eligible. All citations flagged by either reviewer were retrieved for further full-text evaluation to assess their eligibility for quantitative meta-analysis. Only the most recent and comprehensive data were included in the extraction when duplicate publications were encountered by the same author group. Studies were excluded when the intervention comparison did not involve the DS versus VL approach or when both correction approaches were utilized.

Similarly, publications that did not cluster surgical outcomes according to the intervention were excluded. In addition, adult patient studies, single-group cohorts, reviews, and commentaries were excluded. Any disagreements were resolved through reconciliation by a third reviewer (JKK).
Two reviewers (MEC and JMS) independently assessed the risk of bias according to the recommendations of the Cochrane Collaboration for the evaluation of interventional studies (5, 8). No eligible randomized clinical trial (RCT) was identified on the topic, and the included non-RCT comparative studies were assessed using the ROBINS-I tool (8). A funnel plot was generated to assess publication bias.

Data extraction, synthesis, and classification

Study characteristics (study source, study design, sample size, patient age, curvature/hypospadias characteristics, surgical approach, outcome assessed, and follow-up duration) and surgical outcomes were extracted and tabulated by one reviewer (MEC) and counter-verified by two other reviewers (PY and AB). RevMan5 (*Review Manager, Version 5.4, The Cochrane Collaboration, 2020*) program software was used for data analysis and synthesis of the forest and funnel plots. For publications with

incomplete or insufficient data, the event rate (success rate) was extrapolated from available information, such as reported surgical failure or curvature recurrence/persistence.

The primary outcome evaluated in this meta-analysis was the success rate of curvature correction, as assessed in second-stage repair or eventual follow-up. The complication rate was the secondary outcome of the meta-analysis. The reported event rate per intervention group was extrapolated as dichotomous data and pooled as odds ratios (OR) with corresponding 95% confidence intervals (CI). To mitigate methodological variability among the included studies, effect estimates of OR were pooled using the Mantel-Haenszel (M-H) method with a random-effects model to generate an average treatment effect (5, 9, 10).

Assessment of heterogeneity, subgroup analysis, and publication bias

Cochran's $\chi 2$ test was used to assess heterogeneity among the included studies. Considering the small number of studies (and relatively small sample sizes within each) included in the meta-analysis, a *p*-value of <0.10 was applied to detect heterogeneity, while the I^2statistic was utilized to quantify between-study variations, with >40% defined as an assumption of substantial inter-study variability (5). Pre-defined subgroup analyses were performed according to the study population: congenital ventral curvature with or without severe hypospadias and recurrent ventral curvature. The subgroup analysis enables not only the etiology but also the outcome measure differences. A funnel plot generated using RevMan5 was used to determine publication bias. Finally, GRADE criteria were used to assess the certainty of the evidence generated (11).

RESULTS

A total of 1268 records were retrieved from a comprehensive literature search. Subsequently, 72 duplicates were excluded. After an initial screening of 1198 records, 1003 were excluded based on their title and abstract relevance. Finally, a full-text review of 195 articles was performed to determine eligibility; 183 articles were excluded for various reasons, as detailed in the PRISMA flow diagram (Figure 1).

Study characteristics

Twelve non-randomized retrospective comparative studies were included in the meta-analysis (12-23). Seven studies were conducted in the United States (12, 15-17, 20, 21, 23), two in Canada (14, 18), two in Japan (19, 22), and one in Egypt (13). Two studies enrolled patients with congenital ventral penile curvature without hypospadias or with a mild variant of the condition (17, 19). Seven studies included patients with ventral curvature associated with severe hypospadias (13-16, 18, 20, 23), whereas three studies assessed the correction of recurrent ventral curvatures (12, 21, 22). The degree of curvature among the included studies was at least >30.

Eligible studies included patients who had undergone surgery between 1980 and 2018. The patients' ages ranged from 3 months to 21 years. The reported follow-up period for outcome assessment ranged from 6 months to 15 years post-surgery. The DS techniques described included the Nesbit,

Baskin, and Allen-Spence procedures, while the VL techniques included deep transverse incision of tunica albuginea (DTITA)/tunica albuginea incision (TAI) with/without tunica vaginalis flap/graft or dermal/SIS graft. Supplementary Table 1 summarizes the study characteristics in detail.

Study quality

Based on the ROBINS-I risk of bias evaluation of all included studies, the overall assessment was determined to have a high risk of bias. Additionally, bias secondary to confounders was the most prevalent type of bias in the included studies. Supplementary Table 2 details the ROBINS-I evaluation and consensus of the two reviewers.

Treatment effects

Overall, based on the pooled effect estimates from the 12 studies with 430 cases of ventral curvature repair (253 DS, 177 VL), VL repair resulted in a better curvature correction rate than DS repair (OR 4.20, 95%CI 2.11, 8.33) (Figure 2). In the subgroup analysis performed according to curvature etiology, pooled effect estimates from two studies showed that among congenital curvatures with no or mild hypospadias, there was no significant difference in curvature correction between the two approaches (OR 2.99, 95%CI 0.32, 27.57). In the correction of ventral curvature associated with severe hypospadias, based on six studies' pooled effect estimates, VL resulted in a higher rate of curvature correction (OR 3.59, 95%CI 1.25, 10.26). In three studies that assessed recurrent ventral curvature, VL showed a significantly better curvature correction rate than DS (OR 5.70, 95%CI 1.69, 19.21).

Among the five studies that reported the complication rate in each surgical group, the pooled composite complication event rate seemed equivocal for both approaches (OR 0.77, 95%CI 0.27, 2.18) (Figure 3). However, in a study that assessed the complication rate among congenital curvatures with no or mild hypospadias, complications related to urethroplasty (urethral stenosis and urethrocutaneous fistula) were mainly observed in the VL group. Therefore, VL may have resulted in a higher composite complication rate in this group. However, no statistically significant difference between the approaches was noted for the ventral curvature subgroup with severe hypospadias (OR 0.59, 95%CI 0.23, 1.49).

Heterogeneity, publication bias, and GRADE criteria

For overall curvature correction, pooled effect estimates, and subgroup analysis, heterogeneity was not evident based on the chi-squared test, showing a p-value of >0.1 with an inter-study variability of 0%–20%. Similarly, no overall variability was detected for subgroup differences and an I^2 value of 0% justified subgroup analysis of the included studies. Nonetheless, significant overall heterogeneity was noted in assessing composite complications between intervention groups. The chi-square test demonstrated $p = 0.09$ with $I^2 = 50\%$ (Figure 2, 3).

As only a few studies were included, the generated funnel plot could be underpowered; however, this did not suggest the presence of publication bias. The assessed studies were symmetrically plotted across the average OR, with most studies plotting above 1.96 SE (log OR) (Supplementary Figure 4).

Based on the GRADE criteria, the risk of bias was considered serious, with a two-level downgrade on evidence certainty. The assessment of heterogeneity (chi square p value >0.10) and inter-study variability (I square less than 40%) have shown to be not significant for the primary outcome. While for the composite complication, the heterogeneity was explained and improved with subgroup analysis; hence, the domains of inconsistency do not warrant any further downgrading. Although all included studies have directly assessed the surgical success based on the outcome of curvature correction, various ventral lengthening procedures are described even within the study; this has downgraded the certainty of evidence based on the domain of indirectness. With a 30% relative risk reduction and >100 control group event rate, the optimal information size criterion was met with the pooled effect estimates 95% confidence interval has for the primary outcome of success excludes no effect (CI around OR excludes 1.0) precision was determined to be adequate; thus, the domain of imprecision do not render downgrading. Based on the funnel plot generated, no publication bias was suggested, and no further downgrading of the evidence certainty was applied. The domain that could mitigate the risk of bias included a large magnitude of effect, which seemed to be remarkably consistent among patients with severe hypospadias and recurrent curvatures. Thus, the available evidence based on overall GRADE criteria is considered low to very low certainty.

DISCUSSION

Since the first report of congenital penile curvature correction using plication by Reed Nesbit in 1954, dorsal plication has been commonly used to correct congenital penile curvature in patients with and without hypospadias (24, 25). This procedure is widely accepted because of its simplicity and minimal invasiveness. In the preceding decades, there have been various modifications to this technique (26). Owing to the resultant penile shortening and high risk of residual and/or recurrent curvature post-plication in patients with high-grade penile curvature, the approach of VL via corporotomies with and without graft/flap utilization was initiated and has gained some acceptance in recent decades (2, 4). However, the invasiveness of the VL has been widely investigated. Previous surveys have shown that most surgeons prefer dorsal plication to ventral lengthening (25).

This systematic review and meta-analysis synthesized evidence from the literature comparing DS and VL surgical outcomes for penile curvature correction. Concerning surgical outcomes, the pooled effect estimates showed that VL had a higher success rate than DS in the correction of the penile curvature (65.9% vs. 90.0%, respectively; OR 4.20, 95%CI, 2.11, 8.33), while the composite complication rate was comparable to that of DS procedures (40.9 vs. 27.8%, respectively; OR 1.31, 95%CI, 0.46, 3.71). Our findings are consistent with those of another relevant review by Babu and Chandrasekharam (2022), which also found that dorsal plication has a higher risk of recurrence than VL (27). However, our subgroup analysis identifying the significant advantage of VL over DS was only noted for curvature associated with severe hypospadias and recurrent ventral curvatures. While among these subgroups of cases, despite most of the bias of using DS for milder and VL for a more severe degree of curvature, VL still rendered a better correction rate.

This systematic review's literature search process was rooted in the scoping review by Yadav et al. (2022, pending publication), which summarized all studies assessing techniques and comparative outcomes in managing pediatric penile curvature. Our meta-analysis was generated from studies identified through a sensitive search strategy to capture all relevant studies on the topic while restricting studies that reported both the DS and VL approaches and clustered their surgical outcomes accordingly. This enabled a direct comparison meta-analysis to be conducted. In contrast, Babu and Chandrasekharam (2022) reported the prevalence of recurrent curvature for each surgical approach individually and then compared the overall outcome among the Dorsal plication and VL groups, with most included studies reporting only one technique and not both (27). Despite the differences in our methodological approach, both meta-analyses identified similar issues among the available studies, including various outcome measurements and heterogeneity in surgical methodology and reporting. These have restricted the generalizability of the pooled effect estimates, although our study addressed the heterogeneity of population characteristics using subgroup analysis, which improved inter-study variability. However, despite the low inter-study variability and non-evident heterogeneity, we still considered the methodological differences among the included studies. Likewise, the subgroup analysis performed in this meta-analysis addresses the etiological differences of congenital ventral curvature without versus with severe hypospadias versus recurrent curvatures. Thus, giving the readers a better understanding of the effect differences of the contrasting intervention (DS vs. VL) in different etiologies of ventral curvatures in pediatric patients (1-3).

A significant limitation of this systematic review and meta-analysis is that the available literature included all retrospective studies with an inherent methodological constraint of uncontrolled confounders and measurement bias evident in the ROBINS-I assessment. Furthermore, the included studies describe various ventral lengthening procedure techniques. In addition, some have utilized different variable approaches within the study; hence, the downgrading of evidence certainty for indirectness. Despite this limitation, our review contributes considerably to the literature by highlighting the available comparative outcome evidence to inform clinicians of the reported success rate and complications relevant to the procedures in managing penile curvature with or without severe hypospadias. Another critical limitation related to the quality of the available studies included inconsistent measurements of the degree of curvature and a short follow-up period. In most reports, the surgical success of curvature correction was measured during the second-stage procedure and/or a short follow-up period of 1 year; only a few reported long-term follow-up recurrent/persistent curvature, which is clinically and functionally important (28). Hence, surgeons should standardize the measurement using a reliable method, such as the application of technology, to accurately assess the curvature before and after correction (29, 30). Furthermore, long-term follow-up is necessary since recurrent penile curvature after repair affects sexual function (12, 31).

Based on the GRADE criteria for non-randomized comparative studies with a risk of bias downgrading, the certainty of the generated evidence was considered low to very low for recommending one approach over another. Although our study and Babu and Chandrasekharam (2022) similarly

concluded that the VL procedure yields a better outcome yet modest at best. Thus, carefully selecting the appropriate patient population for each approach remains the most crucial management aspect to balance risks and benefits.

CONCLUSIONS

Based on available evidence with low to very low certainty, in the management of selected cases of pediatric penile curvature with severe hypospadias and recurrent ventral penile curvature, VL procedures might render a modestly better success rate than DS. The surgical complication rates were comparable between the two approaches. However, carefully selecting an appropriate patient population for each approach remains the most crucial management aspect to balance risks and benefits. Future studies should be conducted to determine long-term outcomes and standardize the reporting of penile curvature using reliable measurement methods.

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FIGURES AND TABLES

Figure 1. PRISMA 2020 flow diagram for new systematic reviews, which included searches of databases and registers only. Adapted from Page MJ, et al. *BMJ* 2021;372:n71. For more information, visit: http://www.prisma-statement.org/

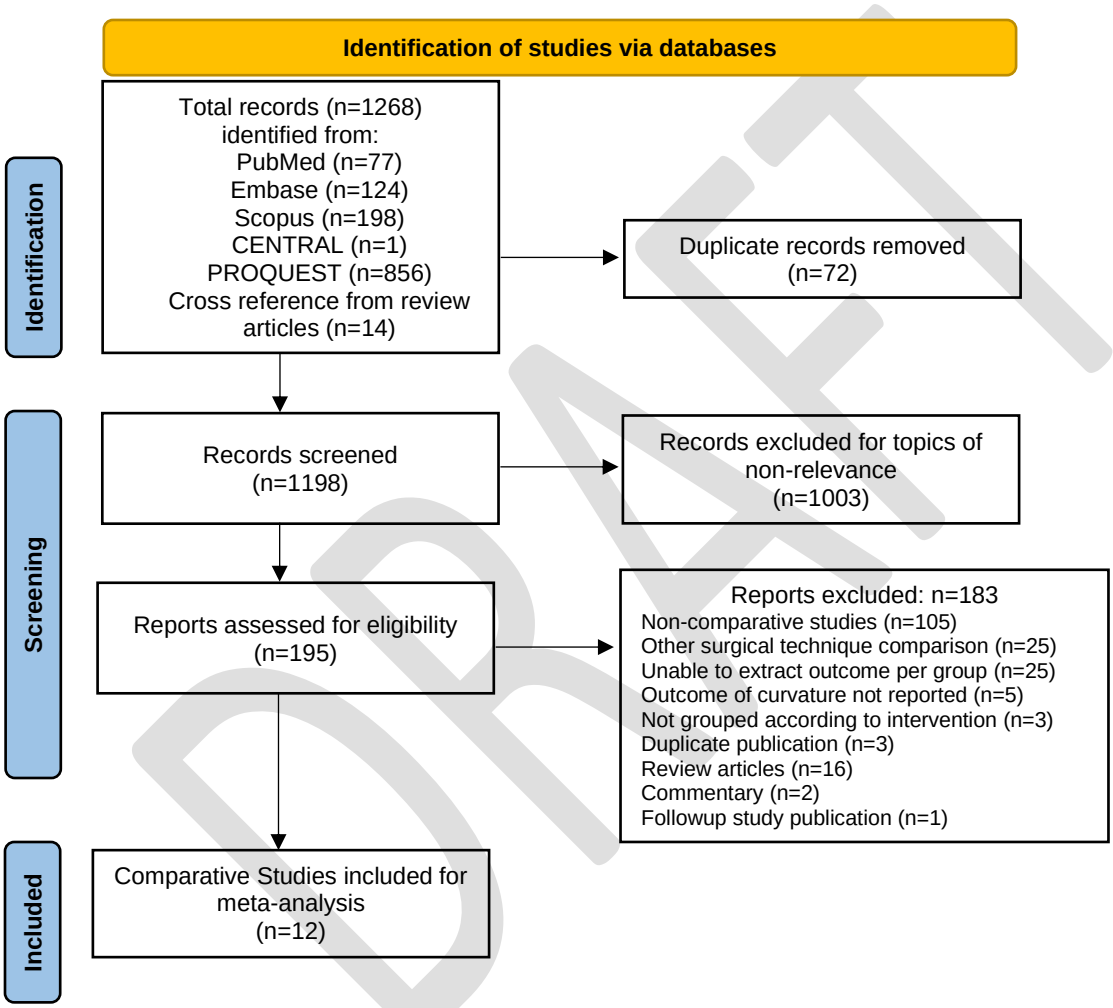

Figure 2. Forest plot pooled effect estimates for outcome of curvature correction rate. Comparison: Dorsal shortening vs. ventral lengthening. Subgroup: Study population (congenital curvature/mild hypospadias/ curvature with severe hypospadias/recurrent penile curvature). Statistical method: Cochran-Mantel-Haenszel method with random-effect model (odds ratio [OR] and 95% confidence interval [CI]).

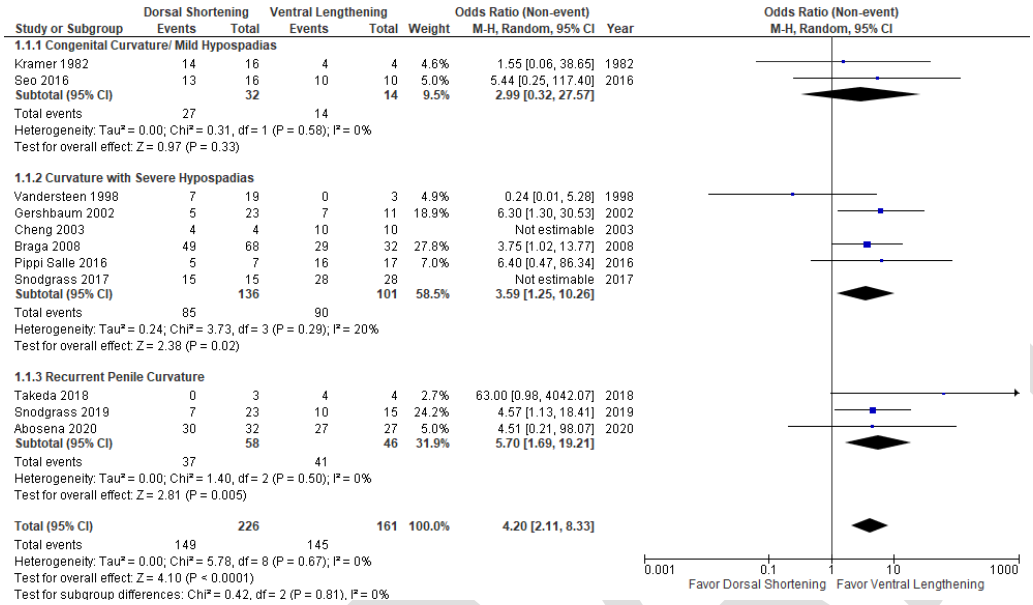

Figure 3. Forest plot pooled effect estimates for outcome of composite complication rate. Comparison: Dorsal shortening vs. ventral lengthening. Subgroup: Study population (congenital curvature/mild hypospadias/curvature with severe hypospadias). Statistical method: Cochran-Mantel-Haenszel method with random-effect model (odds ratio [OR] and 95% confidence interval [CI]).

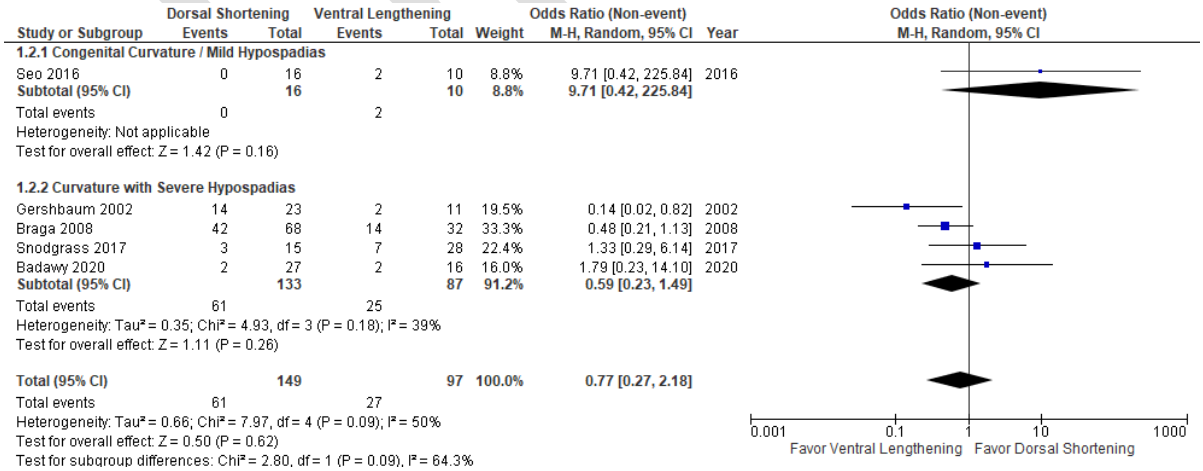

Figure 4. Funnel plot in assessing publication bias.