

PARTIAL VERSUS TOTAL UROGENITAL MOBILIZATION IN UROGENITAL RECONSTRUCTION: A META-ANALYSIS

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ABSTRACT

Objective: Urogenital sinus, cloacal malformation and congenital adrenal hyperplasia are some of the congenital anomalies which comprehensive management should be done to achieve better outcomes. Total Urogenital Mobilization (TUM) and Partial Urogenital Mobilization (PUM) are the most widely used in term of surgical management. However, many researches showed different outcomes of those procedures. This meta-analysis aims to compare the outcome of TUM and PUM in urogenital reconstruction. **Material & Methods:** Studies were collected from five different search engines (PubMed, EBSCO, ProQuest, Science Direct, Google Scholar) with keyword as mentioned in the methods section of this paper. All full text articles were included. Critical appraisal for each study was done using the Oxford Center of Evidence Based Medicine Worksheet for therapy. Data were analyzed by Cochrane's Review Manager 5.3 for charts and plots builder. **Results:** Three studies were found with 81 participants divided into PUM or TUM group. All studies were analyzed by using the Mantel-Haenszel test to produce Forest plot. Overall, urinary incontinence event was more common found in patient who underwent TUM compare with PUM (OR 8.21; 95%CI: 1.1-61.11; $p=0.04$). **Conclusion:** PUM has a better urinary outcome in comparison with TUM. Further study with a better study design, follow-up and standardized evaluation is needed to achieve a better understanding on urogenital reconstruction and management.

Keywords: Partial Urogenital Mobilization, Total Urogenital Mobilization, urogenital reconstruction.

ABSTRAK

Tujuan: Sinus urogenital, malformasi kloaka, dan hiperplasia adrenal kongenital adalah beberapa kelainan bawaan yang membutuhkan manajemen komprehensif untuk mencapai luaran yang lebih baik. Total Urogenital Mobilization (TUM) dan Partial Urogenital Mobilization (PUM) adalah prosedur yang paling banyak digunakan pada tata laksana bedah. Akan tetapi, banyak penelitian menunjukkan luaran yang berbeda dari prosedur tersebut. Meta-analisis ini bertujuan untuk membandingkan luaran dari TUM dan PUM dalam rekonstruksi urogenital. **Bahan & Cara:** Studi dikumpulkan dari lima mesin pencari yang berbeda (PubMed, EBSCO, ProQuest, Science Direct, Google Cendekia) dengan kata kunci sebagaimana disebutkan pada bagian metode dari makalah ini. Semua artikel teks lengkap diikutsertakan. Telaah kritis untuk setiap studi dilakukan dengan menggunakan Oxford Center of Evidence Based Medicine Worksheet untuk terapi. Data dianalisis oleh Cochrane's Review Manager 5.3 untuk pembuat bagan dan plot. **Hasil:** Tiga penelitian ditemukan dengan 81 subjek yang dibagi menjadi kelompok PUM atau TUM. Semua studi dianalisis dengan menggunakan uji Mantel-Haenszel untuk menghasilkan Forest Plot. Secara keseluruhan, kejadian inkontinensia urin lebih sering ditemukan pada pasien yang menjalani TUM dibandingkan dengan PUM (OR 8.21; 95%CI: 1.1-61.11; $p=0.04$). **Simpulan:** PUM memiliki luaran yang lebih baik dibandingkan dengan TUM. Penelitian lebih lanjut dengan desain studi yang lebih baik, follow-up, dan evaluasi sesuai standar diperlukan untuk mencapai pemahaman yang lebih baik tentang rekonstruksi dan tata laksana urogenital.

Kata Kunci: Partial Urogenital Mobilization, Total Urogenital Mobilization, rekonstruksi urogenital.

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INTRODUCTION

Congenital anomalies of the female genital tract can be caused by multifactor, including genetic error or teratologic event during embryonic develop-

ment. Those events may lead to minor or major consequences,¹ some of them are urogenital sinus (UGS), cloacal malformation or congenital adrenal hyperplasia (CAH).^{1,2} The prevalence of congenital malformation were varied across different

populations, starting from 2.46% in Oman, 2.83% in Iran, 4.35% in Taiwan, and 7.92% in United Arab Emirates. In North Indian, out of 20.432 pregnancies, 799 had urogenital congenital anomalies (34 cases of ambiguous genitalia) with an incidence of 39.1 per 1.000 newborns. Moreover, these diseases were more frequently found in stillbirth (12.5%) than in live birth (2.71%).^{3,4}

Beside its incidence, congenital anomalies such as UGS, cloacal malformation or CAH may cause a persistent symptom in their patients. These symptoms can be visible in childhood, puberty or adulthood, ranging from urinary tract infections, post-void dribbling, hematoocolpos, dyspareunia, psychological and psychosexual health, incapacity for sexual intercourse or life-threatening sepsis.^{5,6} Thus, a comprehensive management should be done to achieve better patients' anatomical and functional outcome. In term of surgical management, several techniques were introduced for UGS, starting from the perineal flap in 1964 by Fortunoff et al., pull-through vaginoplasty in 1969 by Hendren and Crawford and prepuce flap in 1989 by Passerini-Gazel.⁵ On the other hand, cloacal malformation patients underwent posterior sagittal anorecto-vagino-urethroplasty.⁶ However, those operations often resulted in complication such as vesicourethral fistulas, urethral and vaginal stenosis, complete vaginal occlusion and urinary incontinence.⁷

In order to minimize the complication and maximize patients' outcome, Pena and Rink et al. published Total Urogenital Mobilization (TUM) and Partial Urogenital Mobilization (PUM) in 1997 and 2006.⁵ TUM and PUM are the most widely used vaginoplasty surgeries.² TUM avoided the separation of urethra from vagina, involved the complete mobilization of both structures, then making separate openings on the perineum.⁷ Meanwhile, PUM does not dissect the common channel completely, but only distal of the pubourethral ligament to limit the possibility of urinary sphincter, clitoris and other structures' injury.⁸ However, many researches shown different urinary continence outcome of those procedures. Some said that TUM might increase the risk of developing urinary incontinence, while others conferred no difference in term of urinary outcome.² Hence, to determine the best option for urogenital reconstruction between TUM and PUM, a meta-analysis was conducted.

OBJECTIVE

Urogenital sinus, cloacal malformation, and congenital adrenal hyperplasia are some of the

congenital anomalies which comprehensive management should be done to achieve better outcomes. Total Urogenital Mobilization (TUM) and Partial Urogenital Mobilization (PUM) are the most widely used in term of surgical management. However, many researches showed different outcomes of those procedures. This meta-analysis aims to compare the outcome of TUM and PUM in urogenital reconstruction.

MATERIAL & METHODS

Studies were collected by using several search engine, those were PubMed, EBSCO, ProQuest, Science Direct, and Google Scholar. The keywords used were “(total OR partial) AND (“urogenital mobilization”). All keywords were searched for their respective MeSH thesaurus. This searching was not limited by date of publication. After the studies were found by searching strategy, those were appraised. Critical appraisal for each study was done using the Oxford Center of Evidence Based Medicine Worksheet for therapy. Data were collected and analyzed by Cochrane's Review Manager 5.3 for charts and plots builder. Our PICO and searching strategy were mentioned in Table 1 and Figure 1.

Table 1. PICO.

Patients	Urogenital anomaly patient
Interventions	Total urogenital mobilization (TUM)
Comparisons	Partial urogenital mobilization (PUM)
Outcome	Urinary continence

This review used all full-text studies which was collected from the search engine mentioned before. Unpublish articles and abstracts only were not included in this study.

The participants of studies in this review were patients with urogenital congenital malformation, like congenital adrenal hyperplasia (CAH), urogenital sinus (UGS), or cloacal malformation patients (CMP). Those criteria were selected because urogenital mobilization vaginoplasty could only be done for those indications.

The intervention was urogenital mobilization vaginoplasty. There were two surgery techniques for vaginoplasty, TUM and PUM. This study aims to compare urinary continence outcome of those techniques. Out of surgery, other treatment modalities were excluded from the analysis.

The outcome of intervention (TUM and PUM) must be followed up at 3-year-old. This age

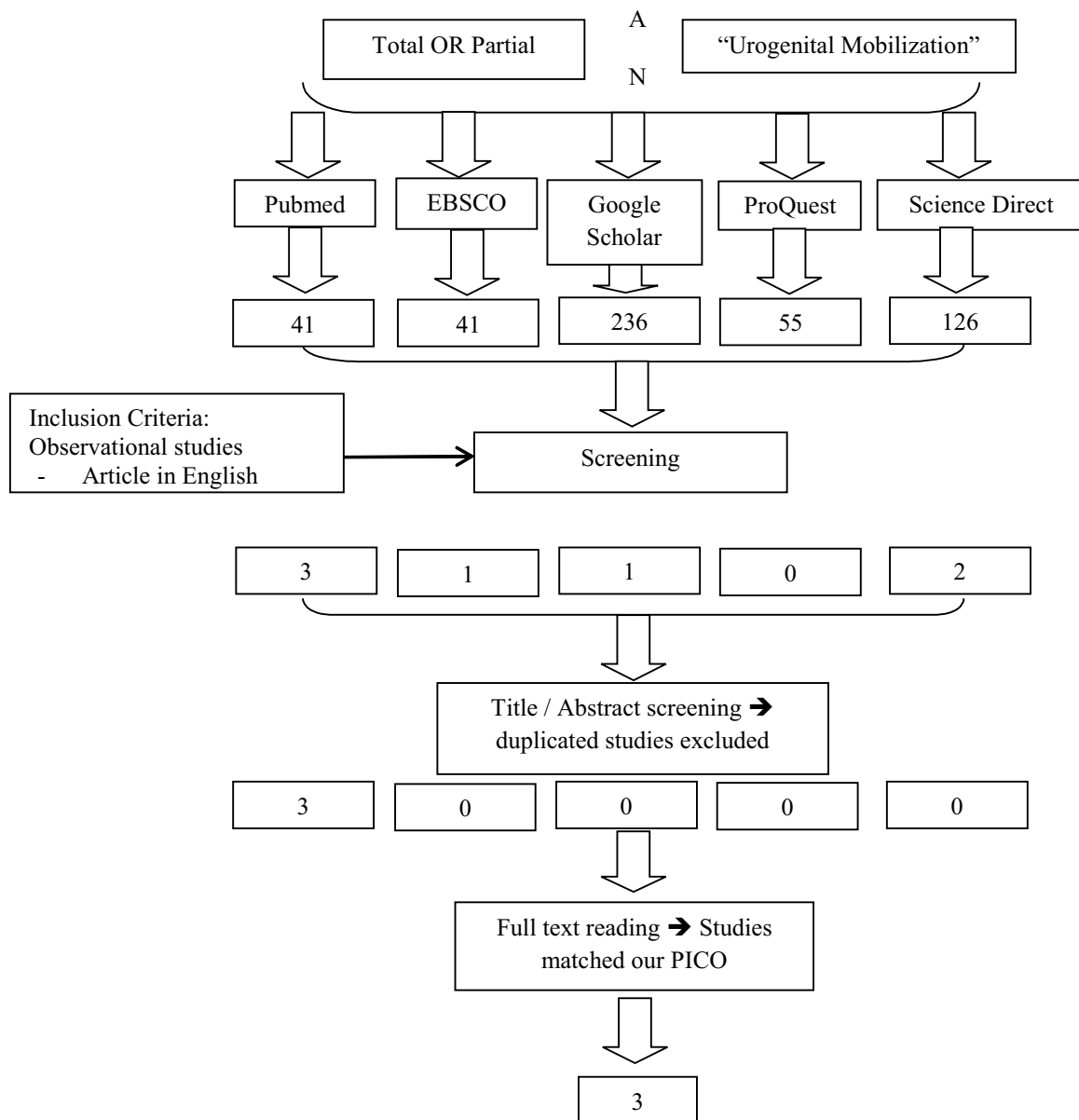


Figure 1. Searching algorithm

was selected because continence post TUM and PUM commonly assessed in this year of life and the time when toilet training should be achieved.

RESULTS

Three studies were comparing the urinary outcome between PUM and TUM. The first study conducted by Leslie et al.⁹ in 2009, involving 44 patients. Out of 44, 18 and 26 patients underwent TUM and PUM with mean follow up of 33 months

and median follow up of 28 months respectively. The median age of going through surgery was 13 months (3 to 174 months) in TUM and 12 months (4-149 months) in PUM group. In terms of outcome, there were 11 patients who experienced incontinence in TUM group, while only one patient in PUM group. The incontinence events were significantly higher in TUM group with OR 39.29 (95% CI 4.3 - 358.85).

The study conducted by Palmer et al.² in 2011 consisted of 25 patients (18 patients TUM and 7 patients PUM) who experience surgery during their

first 1.25 year of life (median, 0.48-17.6), then follow up for 4.41 years (mean, 0.21-12.1). Incontinency was found on one patient in TUM group, but not in PUM. There were three patients (one in TUM and two in PUM group) were not evaluated due to limitation of age (younger than 3-year-old). Based on this study, there was no difference in urinary continence outcome between PUM and TUM (OR 1.00, 95% CI 0.04-28.3).

The latest study found was conducted by Stites et al.¹⁰ in 2016. There were 24 patients participated in this study (TUM: 10 patients; PUM: 5 patients; vaginal pull: through 9 patients). Median age before surgery was 8 months (6-10 months) while duration of follow up was 6 years old (3-10 years).

The result of this study showed that TUM had a higher incidence of incontinency compare to PUM (6 patients to 1 patient in concurrent; OR: 6 (0.04-28.3)).

All studies were analyzed by using the Mantel-Haenszel test to produce Forest plot. The random effects model was used, considering inter-study variability and allowing heterogeneity. These three studies (81 participants) had little heterogeneity ($P=0.18$, $I^2=42\%$), and only one study (Leslie et al.) had significant difference (CI of the OR more than 1). Combined analysis of these studies yields a significant higher risk of urinary incontinence event in patient underwent TUM compare with PUM with OR 8.21 [95% CI 1.1-61.11] and $p=0.04$.

Table 2. Summary of studies included.

	Leslie, et al. ⁹	Palmer, et al. ²	Stites, et al. ¹⁰
Title	Feminizing genital reconstruction in congenital adrenal hyperplasia	Total and Partial Urogenital Mobilization: Focus on Urinary Continence	Urinary continence outcomes following vaginoplasty in patients with congenital adrenal hyperplasia
Year	2009	2012	2016
Location	Texas	Oklahoma	New York
Number of patients	44 patients	25 patients	24 patients
Diagnosis	CAH	CAH,UGS, cloacal anomaly	CAH
Surgery	18 TUM 26 PUM	18 TUM 7 PUM	10 TUM 5 PUM 9 vagina pull through
Median age at surgery	TUM: 13 months (3-174) PUM: 12 months (4-149)	1.25 years (0.48-17.6)	8 months (6-10)
Follow up	median TUM: 33 months PUM: 28 months	mean: 4.41 years (0.21-12.1)	median: 6 years (3-10)
Outcome	TUM: 11 incontinent PUM: 1 incontinent (based on neurological measurement and clinical symptoms)	TUM: 1 patients incontinent PUM: 0 patients incontinence (based on clinical symptoms during follow up) 3 patients less than 3 years and could not be evaluated	TUM: 6 patients incontinence PUM: 1 patients incontinence (based on clinical symptoms during follow up)

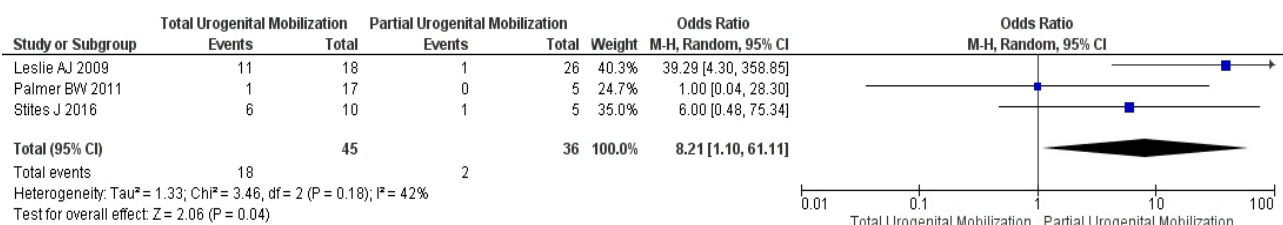


Figure 2. Forest plot of studies

DISCUSSION

UGS is a condition where urethra and vagina are not separated completely during fetal development. Vagina and urethra are joined along bladder neck until perineum, then divided into high and low confluence. The main goal of urogenital reconstruction in this case is to separate urethra and vagina completely, therefore they can work according to their own function.¹¹ In UGS patients with CAH, the main surgery for UGS is still urogenital mobilization to achieve genital function, even though genitoplasty surgeries such as vaginoplasty, labioplasty, and clitoroplasty will also be performed.⁹

There are two types of urogenital mobilization, total and partial urogenital mobilization (TUM and PUM). TUM was introduced by Alberto Pena in 1997. In TUM, circumferential dissection will be made anteriorly and posteriorly to fully mobilize the vagina and urethra. Then, vagina and urethra will be brought closer to the perineum. Anterior dissection is carried through pubourethral ligament. Meanwhile, posterior dissection is done starting from the vagina, allowing the entire UGS complex, like sinus, urethra, and bladder mobilized. Sinus is mobilized closer to perineum and amputated.^{9,11}

Through the development of techniques, anterior dissection which stopped at the proximal of pubourethral ligament (also known as PUM) has been expanded from TUM. Many surgeons believe that PUM can solve urinary incontinence problem in TUM due to the retainment of pubourethral ligament.⁹

Limited studies were comparing the outcome of TUM and PUM in treating UGS. Only

three studies directly compared the urinary continence outcome in patients after TUM vs. PUM. Limitations of these studies include small sample size, retrospective design, heterogeneous nature of pelvic anatomy, and different methods of diagnosing incontinence. Studies conducted by Leslie et al.⁹ and Stites et al.¹⁰ involved patients with CAH, but in Palmer et al.², patients with cloacal anomaly were also included. Patients with CAH and UGS have established perineal musculature and normal neurospinal anatomy, whereas those with cloacal anomaly have an anteriorly rotated pelvic floor and associated lumbosacral spinal malformation. Despite these differences, our combine analysis demonstrate high successful rate among different diagnosis. Only one study, conducted by Leslie et al.⁹ used both neurological measurement and clinical symptoms to diagnose incontinence. The others only based on subjective evaluation by parental interview and clinical assessment, herefore children less than 3 years old cannot be evaluated. Incontinency in Stites et al.¹⁰ consisted of enuresis, voiding postponement, dysfunctional voiding and continuous incontinence.

In combination, patients underwent TUM had higher rate of urinary incontinence compared to patients underwent PUM with OR 8.21 [95% CI 1.1-61.11]. It appears that PUM may be safer from urinary continence standpoint and it is preferred to stop the anterior dissection at the pubourethral ligament wherever possible. Also, it is recommended to use TUM only for patients with very high vaginal confluence.

The limitation of these studies is lack of assessment in outcomes besides urinary continence. Other factors which may be related to urogenital mobilization like cosmetic appearance, satisfactory adult sexual and reproductive function should be

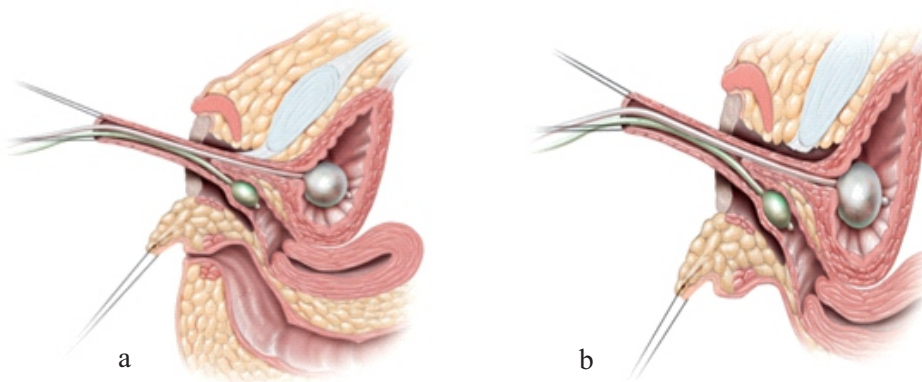


Figure 3. (a) PUM (b) TUM.¹⁰

investigated. Therefore, we recommended long-term follow-up and standardized evaluation in adulthood are needed.

CONCLUSION

Based on our study, PUM has a better urinary outcome in comparison with TUM. However, in patients with high vaginal confluence, TUM is more preferable. In addition, further study with better study design, long term follow-up and standardized evaluation is needed to assess other outcomes such as satisfactory level of sexual, cosmetic, and reproductive function.

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