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Lingual mucosal graft two-stage Bracka technique (



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KEYWORDS

Lingual; Graft; Bracka; Hypospadias; Redo

ABBREVIATIONS

BMG, buccal mucosa graft; LMG, lingual mucosal graft; VAS, visual analogue scale Abstract *Objectives:* To report our initial experience in redo hypospadias repair with a lingual mucosal graft (LMG) using a two-stage Bracka technique.

Patients and methods: This study was prospectively conducted and included 26 patients with hypospadias with failed previous repairs. All the patients had a LMG using a two-stage Bracka technique. In the first stage, the harvested LMG, from the ventro-lateral surface of the tongue, was implanted in a well-prepared vascularised bed in the ventral aspect of the penis. After 6 months, tubularisation of the well-taken graft was completed. Tunica vaginalis or a dartos flap was used as second-layer coverage of the neourethra. Success was defined as acceptable aesthetic and functional outcomes without any additional surgical interventions.

Results: The mean (SD) patient age was 5.15 (1.6) years. The mean (SD) LMG length was 3.82 (0.9) cm and the width was 1.5 (0.5) cm. The mean (SD) number of previous repairs was 2.76 (1.1). The mean (SD) follow-up was 12 (2) months. Donor-site complications included: pain in all patients, with a pain score of >3 on the visual analogue pain scale (0–10) in 10 (38%); and speech problems in 19 (73%). First-stage complications were graft loss (n = 2) and contracture (n = 1). The second stage was completed in 23 patients resulting in the following significant complications: meatal stenosis plus fistula (n = 2), breakdown (n = 1). Successful hypospadias repair was achieved in 77% (20/26) of the patients.

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Conclusion: Lingual mucosa is a reliable and versatile graft material in the armamentarium of two-stage Bracka hypospadias repair with the merits of easy harvesting and minor donor-site complications.

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Introduction

Hypospadias is one of the commonest congenital anomalies with an incidence of up to 1 in 125 live births [1]. One of the most difficult tasks is repair of previously multiple failed hypospadias surgeries, i.e. crippled hypospadias [2–4]. Many surgical techniques have been reported for re-operative hypospadias repair with complications rates ranging from 10% to 57% [3,5–8]. Buccal mucosa graft (BMG) hypospadias repair is the preferred technique when extra genital skin is needed [3,9], with better results obtained with the staged Bracka procedure [4,8–10].

A lingual mucosal graft (LMG) shares the same tissue characteristics and comparable urethroplasty results as a BMG but with easier harvesting and lower donorsite complications [11–18].

In the present study, we aimed to evaluate LMG twostage Bracka redo hypospadias repair for aesthetic and functional outcomes, and donor-site complications.

Patients and methods

This prospective clinical trial was carried out between April 2013 and December 2015. Redo circumcised hypospadaic patients with a scarred fibrotic urethral plate, with or without penile chordee or deficient penile skin, were enrolled in this study. Any patient with oral pathology was excluded from the study. Approval from our Institutional Ethics and Review Committee and informed consent signed by the parents were taken for every case. In all, 26 circumcised redo hypospadaic patients were enrolled in the study (Table 1). A twostage Bracka repair was our chosen surgical technique with a LMG as the tissue substitution for the urethral plate. In the first stage, with the patient under general anaesthesia via a nasotracheal tube, penile degloving and removal of all scarred tissues including the urethral plate was done. Penile straightening was tested by artificial erection (Fig. 1), if residual chordee persisted, a dorsal plication was carried out according to Baskin's modification of the Nesbit procedure [19].



Fig. 1 Graft harvesting from the ventro-lateral surface of the tongue.

Table 1 The patients' characteristics.		
Characteristic		Value
Number of patients		26
Age (at first stage), months, mean (SD, range)		61.8 (19.2, 30–132)
Previous repairs, n, mean (SD, range)		2.76 (1.1, 2–5)
Associated anomalies	Penile chordee, mean (SD, range)	21.25 (8.61, 10-40)
	n = 12	
	Penile rotation, <i>n</i>	6
	Orchidopexy for undescended testis, n	4
Urethral plate defect length, cm, mean (range)		3.08 (1.6-5.7)
LMG length, cm, mean (SD, range)		3.82 (0.9, 2-6.9)
LMG width, cm, mean (SD, range)		1.46 (0.3, 1.2–1.8)
Follow-up period after second stage, months, mean (range)		14.34 (8–22)



Fig. 2 In the first stage, the LMG after being defatted and fenestrated was secured to the prepared bed.

Another surgical team harvested the desired LMG from the ventro-lateral surface of the tongue, such that the length and width taken was $\sim 20\%$ more than the measured urethral plate's defects' length and width. During harvesting, care was taken to avoid injury to the opening of the Wharton's duct and the underlying lingual nerve (Fig. 2). The donor site was closed with running absorbable 4/0 sutures, starting from the posterior end. The graft after being defatted and fenestrated was secured to the prepared bed using interrupted 6/0absorbable sutures. One end of the graft was fixed to the 'V-shaped' dorsally spatulated hypospadaic meatus, whilst the other end was fixed to the tip of a deeply incised glans penis with more fixation sutures between the under surface of the graft and the bed. Finally, the lateral borders of the graft were sutured to the penile skin, which was then covered by a compressive dressing. Patients were discharged on the second postoperative day on oral antibiotic. The first follow-up visit was on the fifth postoperative day where the catheter and dressing were removed, the graft take was judged, penile complications were recorded, and donor-site complications were followed. The subsequent follow-up visits were at 0.5 and 6 months to check graft condition, and donor-site and penile complications.

Donor-site complications (pain, swelling, chewing problems, swallowing problems, oral tightness, speaking

disorders, sensitivity disorders, and difficult tongue protrusion) assessment was done on the second postoperative day, and then at 0.5 and 6 months postoperatively. A visual analogue scale (VAS) was used to assess pain.

Success of the first stage was defined by achieving a straight penis or mild curvature ($< 30^\circ$) and no graft contracture or graft loss.

After ≥ 6 months, the second stage was carried out on those cases with acceptable results in the first stage. Degloving, correction of any residual anomalies, tubularisation of the well-taken graft over a catheter, second-layer coverage of the neourethra by tunica vaginalis or dartos flap, glanuloplasty, and finally skin coverage, were the steps of the second stage of the repair (Fig. 3).

Patients were discharged on the second postoperative day to be followed after 3 days to change the dressing. On the eighth postoperative day, removal of the catheter and dressing, subjective assessment of urine flow and reporting of complications, were done. Patients were instructed to come earlier in cases of wetting or bleeding. Follow-up was done at 0.5, 1.5 and 6 months, then annually. For those patients complaining of a weak stream (subjective observation) of the flow, calibration of the urethra and urethrogram was performed in this order.



Fig. 3 Final intraoperative appearance after the second stage.

Success was defined by having a straight penis with a slit-like meatus at the tip of the glans and no complications that needed any further surgical interventions.

Results

There were 16 posterior hypospadias (penoscrotal type) and 10 middle hypospadias (four mid-penile and six proximal penile). There was no bifid scrotum or penoscrotal transposition. Also, there was no scrotal or perineal hypospadias. Penile chordee was present in 12 cases with a mean (SD, range) curvature of 21.25 (8.6, 10–40)°. After degloving and plate resection the position changed to a more proximal type (20 penoscrotal, four proximal penile and two mid-penile). Penile straightening, determined by artificial erection, was achieved in seven patients by degloving and plate transection (in these cases penile chordee was due to skin tethering and scared fibrotic plate) and in the other five patients dorsal plication was needed. The mean (SD) time of LMG harvesting was 18.2 (2.9) min.

First-stage complications included: infections (n = 3), penile oedema (n = 6), skin ecchymosis (n = 2), mild residual chordee (n = 2), graft loss (n = 2), and graft contracture of > 20% (n = 1). Additional surgical interventions in the form of re-grafting with BMG were only needed in cases with graft loss and contracture, which was considered as failure (11.5%).

Donor-site complications were mild in nature and disappeared gradually with time, with three long-term complications in two patients. There was no bleeding or haematoma formation. Pain at the donor site was present in all patients, 18 (69%) of them required analgesia (Table 2). Three of our patients complained of dressing wetting and thus this was changed before the eighth postoperative day.

In all, 23 cases were suitable for tubularisation in the second stage, which resulted in 14 complications in seven patients that included: infection (n = 2), oedema (n = 4),

Table 2Complications.			
	Second postoperative day	0.5 months	6 months
VAS score, mean (range)	3.5 (1-9)	1.3 (0-6)	0.15 (0-2)
N (%):			
VAS score of > 3	10 (38.46)	3 (11.5)	0
Analgesia required	18 (69.2)	0	0
Swelling in graft site	7 (26.9)	3 (11.5)	0
Problems with chewing	17 (65.38)	4 (15.3)	0
Problems with swallowing	10 (38.46)	2 (7.69)	0
Oral tightness	0	0	0
Speaking disorders	19 (73.03)	8 (30.8)	2 (7.69)
Sensitivity disorders	12 (46.1)	5 (19.2)	0
Difficult tongue protrusion	4 (15.3)	3 (11.5)	1 (0.038)
More than two complications	15 (57.69)	7 (26.9)	1 (0.038)

skin ecchymosis (n = 1), bleeding (n = 1), fistulae (n = 3), repair breakdown (n = 1), and meatal stenosis (n = 2). Most of these complications responded to conservative management. Three of the seven patients with complications needed re-operation. There were two cases with fistulae and one case with repair breakdown resulting in a success rate of 20/23 (87%) for the second stage. In the third case with fistula, calibration and dilatation were done with bougie, resulting in spontaneous fistula closure. Thus, the overall success rate in our series was 20/26 (77%). The mean (SD) follow-up was 12 (2) months.

Discussion

The goals of hypospadias repair are a slit-like meatus at the tip of the glans and a straight penis that allows a forward directed urinary stream, a urethra with adequate calibre, and normal coitus, which can be achieved in most cases by a one-stage repair and as such is considered the standard of care in current practice, but this is difficult to achieve in cases with multiple failed previous repairs [3,4]. Especially as these difficult cases often present with worse deformities compared with the primary congenital anomalies [3].

The Bracka two-stage hypospadias repair's versatility and reliability is recognised, with acceptable success rates in both primary and redo procedures, especially when BMG is used [8,9,20]. Complete graft take, more reliable revascularisation, and exclusion of graft contracture before the second-stage tubularisation are the main advantages of this technique, with better cosmetic and functional results than after single-stage repair [8– 10]. A LMG, as oral mucosa, can be used for urethral reconstruction with high success rates and mild donorsite complications [11–15].

In a comparative study between LMG and BMG urethroplasty, Chauhan et al. [21] found comparable urethroplasty results but there were fewer and less morbid donor-site complications with LMG in the short-and long-term.

In another study by Kumar et al. [12], although early donor-site complications were reported more with LMG than with BMG, the long-term complications (tightness, discomfort, and numbness) were only present in the BMG group. Similar results were also reported by Pal et al. [18], with slurred speech present only with LMG in the early postoperative period, which disappeared on longer follow-up where salivatory changes, tightness, pain at the graft site and numbness were present only in the BMG group. Also, a shorter harvesting time of LMG vs BMG (25 vs 38 min) was reported, being an additional advantage of LMG, which was also reported by Sharma et al. [13] with a shorter unilateral LMG harvesting time than BMG harvesting time (13.2 vs 16 min).

Maarouf et al. [22], found LMG to be superior to BMG in terms of mouth tightness, oral discomfort and numbness, although it is inferior to it in terms of slurred speech and tongue protrusion.

Lumen et al. [23] reported no significant difference in postoperative pain, need for analgesia, or hospital stay between LMG and BMG donor-site morbidity. Also, although there were significant differences in favour of BMG at the third postoperative day for eating and drinking, speaking disorders, and dysgeusia, by the second week, significance only persisted for speaking disorders with the appearance of a new significant difference in oral tightness with BMG vs LMG (41.4% vs 6.9%, P = 0.005). At the 6-month follow-up, there were no significant differences between the two groups with only a difference tending towards significance in oral tightness in favour of LMG (P = 0.052).

In the present study, 26 patients with failed previous hypospadias repairs underwent a LMG two-stage Bracka technique repair. The lengths and widths of the LMGs in our present study were nearly equal to those of Simonato et al. [11], shorter than those of Simonato et al. [14] and much shorter than those of Kumar et al. [16]. The difference between the LMG size in our present study and others may be because our study was conducted on hypospadias cases in children aged < 6 years; however, all the other LMG studies mentioned were for urethral strictures in adults and not for hypospadias. Also Kumar et al. [16] harvested the LMG from the entire length of the lateral surface of the tongue as one unit reaching \sim 16 cm in length.

For donor-site complications, in our present study they were mild in nature and most of them disappeared by the second week postoperatively, with only three long-term complications (two speech disorders and one difficult tongue protrusion) after 6 months.

All patients in our present study had pain at the time of discharge (second postoperative day), which was also reported by Sharma et al. [13], but with a lesser mean VAS pain score (3.5 vs 7.4). Pain occurrence was less in the studies of Kumar et al. [12,16] and Xu et al. [17]. In the present study, 10 patients (38%) had a VAS pain score of > 3 and 18 (69%) needed analgesia to relieve pain.

Speaking disorders occurred in 19 patients (73%) being the commonest complication on the second postoperative day, which decreased to eight patients (30%) after 2 weeks. This is less than the 100% reported by Simonato et al. [14] and Sharma et al. [13], comparable to that of Xu et al. [17] (69%) when bilateral harvesting was done, and more than that of Kumar et al. [12,16] (22%, 24%) and Xu et al. [17] (22%) when unilateral harvesting was done. Two cases of speech disorders remained after 6 months as long-term complications, which also occurred in 17% of patients in the Xu et al. [17] study and 20% of the Sharma et al. [13] patients, but in none in the studies of Simonato et al. [14] and Kumar et al. [12,16]. In the present study, chewing and swallowing difficulties occurred in 17 (65%) and 10 (38%) patients, respectively, which is far less than that reported by Sharma et al. [13], which was a 100% for both. Difficult tongue protrusion occurred in four patients (15%) on the second postoperative day, which decreased to three (12%) by the second week, with one persisting as a long-term complication; whereas in the study of Sharma et al. [13], there were five cases (five of 15) on the second postoperative day, with two of them persisting as a longterm complication.

Chauhan et al. [21] reported that difficult chewing and altered sensation persisted more in their BMG group up to the third postoperative month. Similar results were reported by Pal et al. [18], showing lower donor-site complications in favour of LMG vs BMG, although complications occurred more with LMG in the early postoperative period these disappeared rapidly with persistence of complications only in the BMG group.

More than two complications in the same patient occurred in 15 (58%) of our present patients at the second postoperative day, which decreased to seven (27%) by the second week, and to one (0.04%) at 6 months. Lumen et al. [23] reported that two or more complications occurred in eight (27.6%) and five (17.2%) patients at the 6-month follow-up treated with BMG and LMG, respectively (P = 0.345).

In the present study, re-grafting was done using BMG and was successful after one procedure. We avoided re-harvesting form the other side of the tongue so as not to increase donor-site complications, which may be a limitation of LMG. Our re-grafting rate (11.5%) is similar to that reported by Bracka [9] (10%) in a series of > 121 patients and more than that of Nerli et al. [24] (two of 21, 9.5%) and Johnson et al. [25] (one of 17, 5.9%).

The second-stage success rate (20/23, 87%) in our present study is better than that of Sripathi et al. [26] (50%) and Johnson et al. [25] (64%), but lower than that of Nerli et al. [24] (90.5%), Bracka [9] (90%), and Gill and Hameed [27], which was >90%.

Fistula in our present study occurred in three of 23 cases (13%), the two cases (9%) with meatal stenosis, needed formal fistula closure and meatoplasty. This fistula rate is similar to that of Nelson et al. [28] (11.6%) and Sripathi et al. [26] (10%), but higher than that of Amukele et al. [29] (0%), Nerli et al. [24] (4.7%), Fichtner et al. [30] (4.08%), and Gill and Hameed [27] (9%). But, all of those studies used a BMG, we could not find any studies reporting the use of LMG for Bracka two-stage repair in redo hypospadias.

In our present study, failed cases, either in the first- or second-stage, all were complicated with infection, which is considered one of the greatest enemies of graft surgery. The overall success rate was 20/26 (77%) over a mean (SD, range) of follow-up of 12 (2, 8–22) months. Extended follow-up is needed, as complications can still occur over time.

Conflict of interest

None.

Financial disclosure

None.

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