Outcome of severe hypospadias repair using three different techniques

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Received 9 October 2008; accepted 17 December 2008
Available online 7 February 2009

Abstract

Objective: To compare the outcomes of three different urethroplasty techniques (onlay, buccal mucosa, Koyanagi type I) used in the reconstruction of severe hypospadias. Patients and methods: Over 10 years (1997–2007), 300 severe hypospadias cases were treated with a mean follow up of 2 years (1–105 months); 203 were operated by the same surgeon of whom 184 completed follow up. Three main techniques were used according to the quality of the urethral plate: onlay urethroplasty (133), buccal graft urethroplasty (25) and Koyanagi type I (26). The mean age at surgery was 36 months (8–298); 76 required preoperative androgen stimulation (onlay 37, buccal 11, Koyanagi 26); 18 required a corporoplasty to straighten the penis (onlay 13, buccal 3, Koyanagi 2). Results: Thirty-eight onlay (28.5%); 14 buccal (56%); 16 Koyanagi (61.5%) urethroplasties had a complication. The fistula rate was 15% for the onlay group; 32% for the buccal mucosa group; 19.2% for the Koyanagi cases. The dehiscence rate was, respectively, 11.3%, 20% and 42.3%. The stricture rate was, respectively, 1.5%, 20% and 34.6%. Urethrocele was found in seven Koyanagi patients. Final functional and cosmetic results were satisfactory in 126/133 (94.7%) onlay, 20/25 (80%) buccal and 14/26 Koyanagi (53.8%) urethroplasties. Primary cases had better results (89%) than redo cases (75.9%). Patients submitted to preoperative androgen stimulation developed more complications (onlay: 40.5% vs 23.9%; buccal: 70% vs 43.7%). Conclusion: Two striking results are the low number of severe hypospadias cases requiring an additional corporoplasty, and the increased complication rate found in androgen-stimulated patients. The excellent results of the onlay procedure could be related to the use of dorsal preputial tissue, which in hypospadias is characterized by a well-balanced protein platform compared to the ventral tissues.

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Introduction

Reconstruction of severe hypospadias remains a challenge for the Pediatric Urologist as most procedures described are characterized by a significant number of complications and redo operations. A literature review shows that in severe hypospadias the most commonly used procedures are: the Asopa–Duckett tube [1], the onlay island flap urethroplasty [2], the buccal mucosa graft urethroplasty [3], the Koyanagi procedure [4], the tubularized incised plate (TIP) procedure [5] and the Cloutier-Bracka two-stage procedure [6,7]. Long-term outcomes of these procedures are poorly documented.

Among 300 severe hypospadias cases operated in the same institution over the last 10 years, we selected those operated upon by the same surgeon using three main techniques: the onlay island flap urethroplasty, the buccal graft urethroplasty and the Koyanagi type I procedure. Results were reviewed and compared to those published. Potential effects of hormonal stimulation on the healing process are discussed.

Patients and methods

The medical records were reviewed retrospectively and selected hypospadias cases were those with a proximal division of the corpus spongiosum found after degloving the penis and marked hypoplasia of the ventral tissues (Figs. 1 and 2) associated with penile curvature. All were operated on by the same surgeon (PM) in the same institution following the same protocol over a period of 10 years (1997–2007). Of 204, 15 were excluded from the study because they were lost from view after surgery, and the five Bracka procedures [6,7] performed were excluded because of our insufficient number of cases and experience with this procedure. Hence, a total of 184 cases had a documented mean follow up of 24 months (1–105).

The choice of technique was dependent upon the quality of the urethral plate. For primary hypospadias with a healthy urethral plate, the first choice was the onlay...
island flap urethroplasty [8] (Fig. 3); for those with no foreskin available (mainly redo cases), the buccal graft urethroplasty [9] (Fig. 4) was selected; and for those with a perineal division of the corpus spongiosum, the Koyanagi type I [4] (Fig. 5) procedure was favored.

The surgical procedures followed the same steps as described elsewhere [10]. They were all performed with a tourniquet and bipolar diathermy coagulation was seldom used. All patients received the same postoperative protocol, including a daisy dressing (Fig. 6) using sulfadiazine and hyaluronate cream (Ialuset plus®), a silicone mesh (Mepitel®), and a Kling bandage maintained by a strip of Elastoplast®. The dressing was changed on day 4 postsurgery and removed on day 10 along with the Vygon 8 silastic catheter. Children under 2 years of age had a double nappy whereas older children had a collecting bag. All received an intravenous injection of cephalosporin at the time of surgery followed by an oral sulphametoxazol–trimethoprim cover until the catheter was removed. Preoperative endocrine and genetic work ups and hormonal stimulation were indicated according to the size of the penis, the severity of the hypoplasia of the ventral radius of the penis, and a history of previous surgery.

Results

The outcome of 184 severe hypospadias cases operated by the same surgeon in the same institution from 1997 to 2007 using the onlay urethroplasty, buccal mucosa graft urethroplasty and Koyanagi procedures was analysed. Patients were separated according to the technique performed (Table 1) and the number of previous procedures (Table 2).

One hundred and fifty-five cases were primary repairs whereas 29 patients had undergone one or several previous surgical procedures (Table 1). The mean age at surgery in all 184 children was 36 months (8–298); 24 months (8–96) in the onlay group, 100 months (20–298) in the buccal mucosa group and 23 months (9–40) in Koyanagi group. Patients received preoperative hormonal stimulation when the penis was particularly small or hypoplastic, or in the case of redo surgery. Three main types of androgen stimulation were performed: human chorionic gonadotrophin (beta-hCG) at a dose of 1500 IU every other day for 12 days; intramuscular testosterone (Androtardyl®) at a dose of 100 mg/m²; and topical dihydrotestosterone (Andractim®), one application per day on the ventral aspect of the penis for 2 months. Only 13 patients with persistent corporeal curvature after complete penile dissection required a tunica albuginea plication corporoplasty [11] in the onlay group. Only two received a corporoplasty in the Koyanagi group.

The mean follow up of the 184 children was 24 months (1–105) with some variation between the three groups.

<table>
<thead>
<tr>
<th>Type of repair</th>
<th>Primary repair</th>
<th>Previous failed procedures</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Onlay</td>
<td>126 (94.7%)</td>
<td>7 (5.3%)</td>
<td>133</td>
</tr>
<tr>
<td>Buccal mucosa</td>
<td>3 (12%)</td>
<td>22 (88%)</td>
<td>25</td>
</tr>
<tr>
<td>Koyanagi</td>
<td>26 (100%)</td>
<td>0</td>
<td>26</td>
</tr>
<tr>
<td>Total</td>
<td>155 (84.2%)</td>
<td>29 (15.8%)</td>
<td>184</td>
</tr>
</tbody>
</table>
One hundred and four postoperative complications were recorded in 68 patients (36.9%): 38 with the onlay (28.5%), 14 with the buccal mucosa (56%) and 16 with the Koyanagi (61.5%) procedure (Tables 3 and 4). Fistula was found in 15% of onlay, 32% of buccal mucosa and 19.2% of Koyanagi procedures; dehiscence in 11.3%, 20% and 42.3%; and stricture in 1.5%, 20% and 34.6%, respectively. Seven Koyanagi procedure cases (26.9%) developed an urethrocele (Table 4). Other complications included: skin irregularities (3), balanitis xerotica obliterans (2), residual chordee (1), dysuria/vesicourethral dyssynergia (2) and meatal retraction (1) with the onlay technique; and concealed penis (2) and meatal retraction (1) with the buccal mucosa technique.

Final functional and cosmetic results were satisfactory in: 126/133 (94.7%), 20/25 (80%) and 14/26 (53.8%) patients submitted to the onlay, buccal mucosa and Koyanagi techniques, respectively, regardless of the number of procedures; and 138/155 (89%) of patients undergoing primary repair and 22/29 (75.9%) of patients with previous surgery (Table 5). There are 24 patients awaiting another operation: six with partial dehiscence, seven with fistula and 11 with penile or perineal urethrostomy (Table 6).

Out of 73 patients who received preoperative androgen stimulation, 37 were onlay procedures, 10 buccal mucosa and 26 Koyanagi procedures. Out of 37 stimulated onlay patients, 23 received hCG and 26 systemic testosterone. Of the 10 stimulated buccal mucosa patients, seven received hCG, seven systemic testosterone and two received topical dihydrotestosterone. Comparison of outcomes between stimulated and non-stimulated patients showed a significant difference between the onlay and buccal mucosa patients (Table 7).

### Discussion

**Anatomical considerations and the choice of techniques for urethroplasty in severe hypospadias**

The severity of hypospadias can only be established with certainty at the time of surgery when the penis is degloved and when the division of the corpus spongiosum is clearly identified. The position of the ectopic urethral meatus is a poor criterion to define the severity of a hypospadias case, as quite frequently the division of the spongiosum is significantly more proximal than the meatus. Once this is recognized, the number of severe hypospadias cases, i.e., hypospadias with a division of the corpus spongiosum sitting proximal to the penile midshaft, associated with a hypoplasia of the ventral tissues is more frequent than classically reported in textbooks [12]. It is a well recognized fact that surgery for severe hypospadias leads to more complications than more distal hypospadias, whichever technique is chosen, and is still one of the most challenging surgical procedures facing the hypospadiologist [13].

The choice of procedure is very much surgeon dependent. In our experience, the key parameter in selecting the technique of urethroplasty is the quality of the urethral plate. This can only be established once the penile dissection has been fully completed, and this is the reason why all technical options to reconstruct the missing urethra should remain available until the final choice is made. If the urethral plate is healthy and if the inner prepuce is available, our first choice is the onlay island flap urethroplasty. If the child already underwent an operation which failed and if the prepuce is no longer available, the buccal graft urethroplasty is favored, knowing that free graft urethroplasty would be expected to heal with a better success rate.
has less good results than when using a pediculized graft. Finally, in the most severe forms of hypospadias, i.e., with a perineal division of the spongiosum and major hypoplasia of the ventral tissues, we choose the Koyanagi procedure.

Outcome of reported severe hypospadias repairs

Baskin et al. [14], in a series of 374 onlay island flap cases, reported that only 50 patients (13%) still had a significant chordee after degloving the penis. This result was confirmed in our series, where only 18/184 (9.8%) required chordee correction after fully degloving the penile skin shaft in primary hypospadias repair. Baskin et al. stated that the urethral plate must be preserved if possible, even in proximal forms of hypospadias, and that it is not usually the cause of penile curvature. They published a very low fistula rate of 6% with the onlay technique. The incidence of fistula after onlay procedure in our study was 15% which is comparable to several other publications. Wallis et al. published a fistula rate of 20% in patients operated using the onlay technique, in an article comparing onlay island flap urethroplasty with TIP [15].

Other recent publications reported overall complication rates of 31% [16], 22.5% [17] and 45% [14] with the onlay island flap technique, which are similar to our series where a 28.5% complication rate was found. Markiewicz et al. [18] reported an 80% success rate with the buccal graft urethroplasty and the free preputial graft which is comparable to our final results (80%).

There is very little long-term data on the Koyanagi procedure. The Necker team in Paris has achieved results (unpublished data) with the modified Koyanagi (Hayashi) [19] which are very similar to our results. This modification may improve the blood supply of the distal skin flaps. We have now started to use this modification and will report on it in due course. With the type I procedure, the main drawbacks were represented in our series by the difference in compliance between the native urethra and the long reconstructed urethra. In addition, the distal granular reconstructed urethra tends to narrow down with time increasing the urethral flow pressure, which facilitates the development of an urethrocele. Better backing of the reconstructed urethra with the surrounding tissues and a large distal urethra may contribute to a better outcome. A striking result of this study is the very low number of Koyanagi cases (2) which required a complementary corporoplasty to straighten the penis. This finding goes against what we [8] and others [20] published in the past, when we stated that the urethral plate is rarely the cause of the chordee even in severe hypospadias. These 26 Koyanagi cases all had a division of the plate and full dissection of all tissues lying beyond the division of the corpus spongiosum, not only on the ventral surface of the corpora cavernosa but also on each side of the penis and on the dorsum. The concept of preserving the urethral plate in perineal hypospadias needs to be challenged as the degree of hypoplasia of the ventral tissues is of a different magnitude in these cases [12].

Alternative techniques like the Cloutier-Bracka two-stage repair [6,7] are commonly used by other teams, but our experience was limited to five cases which was insufficient to allow a comparison. According to Bracka’s experience of 600 cases, fistulae were found in 5.7% (3% for primary repair and 10.5% for salvage surgery) [6]. In a recent article by the same author, comparing different techniques with one- and two-stage repairs, it is stated that the Bracka procedure remains an ideal and versatile solution when circumferential urethroplasty is required, whether in primary or reoperative hypospadias [21].

Table 4 Types of complication.

<table>
<thead>
<tr>
<th>Complication</th>
<th>Onlay</th>
<th>Buccal mucosa</th>
<th>Koyanagi</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Primary</td>
<td>Primary</td>
<td>Primary</td>
<td></td>
</tr>
<tr>
<td>Fistula</td>
<td>18</td>
<td>2</td>
<td>13</td>
<td>33</td>
</tr>
<tr>
<td>Stricture</td>
<td>2</td>
<td>0</td>
<td>5</td>
<td>9</td>
</tr>
<tr>
<td>Dehiscence</td>
<td>13</td>
<td>2</td>
<td>5</td>
<td>16</td>
</tr>
<tr>
<td>Mucosal prolapse</td>
<td>4</td>
<td>0</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Urethrocele</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>Others</td>
<td>9</td>
<td>1</td>
<td>2</td>
<td>12</td>
</tr>
<tr>
<td>Total</td>
<td>47</td>
<td>6</td>
<td>15</td>
<td>104</td>
</tr>
</tbody>
</table>

Table 5 Results of operation.

<table>
<thead>
<tr>
<th>Operation</th>
<th>Onlay</th>
<th>Buccal mucosa</th>
<th>Koyanagi</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Initial good results</td>
<td>99</td>
<td>0</td>
<td>10</td>
<td>124</td>
</tr>
<tr>
<td>Reoperated patients</td>
<td>24</td>
<td>3</td>
<td>9</td>
<td>38</td>
</tr>
<tr>
<td>Good results after reoperation</td>
<td>22</td>
<td>3</td>
<td>5</td>
<td>36</td>
</tr>
<tr>
<td>Final good results</td>
<td>121</td>
<td>5</td>
<td>14</td>
<td>160</td>
</tr>
<tr>
<td>Cases waiting for reoperation</td>
<td>5</td>
<td>0</td>
<td>5</td>
<td>12</td>
</tr>
</tbody>
</table>

Table 6 Cases awaiting surgery.

<table>
<thead>
<tr>
<th>Operation</th>
<th>Onlay</th>
<th>Buccal mucosa</th>
<th>Koyanagi</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fistula</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>Urethroscopy</td>
<td>1</td>
<td>0</td>
<td>2</td>
<td>11</td>
</tr>
<tr>
<td>Dehiscence</td>
<td>4</td>
<td>1</td>
<td>1</td>
<td>6</td>
</tr>
<tr>
<td>Urethrocele</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>5</td>
<td>2</td>
<td>5</td>
<td>12</td>
</tr>
</tbody>
</table>
TIP is a more recent technique that is considered by some as simple with a low complication rate when used for distal repairs [22]. This technique can also be used for proximal forms of hypospadias. Snodgrass and Lorenzo published their experience with 33 consecutive patients undergoing the TIP procedure, with complications in 11 (33%) patients (21% developed fistulae). The authors considered the technique to be a reasonable option for proximal forms without significant chordee and with a supple appearance of the plate [23]. TIP is considered by the same authors as also potentially usable for hypospadias redo operations [24]. In our institution we do not have any experience of this technique in severe hypospadias.

The tissue factor in the outcome of hypospadias surgery

Complications are not only related to the technique chosen and the surgeon’s experience but also to the intrinsic qualities of the tissues forming the hypospadiac penis. The degree of hypoplasia of the ventral tissues in the most severe forms of hypospadias and the presence of scarred tissues after failed surgery are significant factors influencing the surgical outcome. The quality of healing is a major concern in hypospadias surgery. Various attempts have been made to boost the factors involved in the healing process [25,26]. Among them, androgen stimulation of the hypospadiac penis prior to surgery is the most commonly used, not only to make the penis bigger but also to increase the tissular blood supply and possibly to boost the healing performance of the penile tissues. Koff and Jayanthi reported the use of hCG in 12 severe hypospadias cases prior to surgery and noticed a downgrade of the severity of the condition after 5 weeks of treatment. They showed that in all cases chordee decreased and penile length increased mostly proximal to the urethral meatus, which significantly moved the meatus distally and facilitated surgical treatment [27]. Another striking result of the analysis of our series is that we found that patients who received preoperative androgen stimulation had significantly more complications (46.8%) than those who did not receive any stimulation (26.8%), in the onlay group (39.5% vs 24.2%) and in the buccal mucosa group (70% vs 43.7%) (Table 7). One could argue that the patients who received hormonal stimulation were the most severe cases, although we noticed that all patients who received this stimulation had a much better trophicity of the tissues forming the ventral radius of the penis. We are also puzzled by statements made by dermatologists regarding the endocrine aspect of the skin healing process. Gilliver et al. published a paper about hormonal regulation and the contrasting contributions of sex steroid hormones to the regulation of cutaneous wound healing. According to these authors, oestrogens can modulate the inflammatory reaction, accelerating re-epithelialization, and stimulating angiogenesis and wound contraction. Androgens, on the other hand, have been identified as repressors of cutaneous repair, retarding the healing process and increasing inflammation [28]. This raises the question of the choice of stimulation prior to hypospadias surgery, knowing that it is most unlikely that surgery alone can fully correct these severe forms of hypospadias.

A preliminary study performed on the protein content of hypospadiac tissues demonstrated that the balance between structuring (cadherin E and claudin 1) and destructuring (metalloproteinase 2) proteins differs strongly between the dorsum and the ventrum of the hypospadiac penis compared to controls [29]. This finding could explain the better results obtained with the onlay island flap urethroplasty, where dorsal skin with normal protein balance is transferred ventrally, potentially leading to a better quality of tissue healing.

Conclusions

Our series and others confirm that the onlay urethroplasty is a major advance in the reconstruction of severe hypospadias. It is possible that the modification of the Koyanagi procedure published by Hayashi [19] achieves better results although there is no evidence for this yet. Buccal mucosa is an essential substitutive tissue for the urethra, although one cannot expect straightforward good results in most cases with free graft urethroplasty. Parents should, therefore, be clearly informed that surgery of severe hypospadias often requires several steps. Finally, surgery is only one part of the armamentarium available to treat hypospadias. Further studies are needed to determine which form of stimulation is the most appropriate to boost tissue healing in hypospadias surgery to compensate for the possible intrinsic protein imbalance.

Conflict of interest/funding

None.

References


