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Haytham Badawy & Ahmed Fahmy

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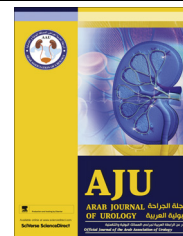
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PEDIATRIC UROLOGY

REVIEW

Single- vs. multi-stage repair of proximal hypospadias: The dilemma continues

Haytham Badawy *, Ahmed Fahmy

Department of Urology, University of Alexandria, Alexandria, Egypt

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KEYWORDS

Single-stage;
Multi-stage;
Hypospadias;
Repair;
Complications

ABBREVIATION

TIP, tubularised
incised-plate

Abstract Introduction: The surgical reconstruction of distal penile hypospadias in a single stage is the standard practice for managing anterior hypospadias. Unfortunately, it is not simple to extrapolate the same principle to proximal hypospadias. There is no consensus among hypospadiologists about whether a single- or multi-stage operation is the optimal treatment for proximal hypospadias. In this review, we assess the currently reported outcomes and complications of both techniques in proximal hypospadias repair.

Methods: We searched Medline, Pubmed, Scopus and Ovid for publications in the last 10 years (2002–2012) for relevant articles, using the terms ‘proximal hypospadias’, ‘posterior hypospadias’ ‘single stage’, ‘multiple stage’, and ‘complications’. Articles retrieved were analysed according to the technique of repair, follow-up, complications, success rate, number of included children, and re-operative rate.

Results and conclusions: The reported complications in both techniques were similar, including mostly minor complications in the form of fistula, meatal stenosis, partial glans dehiscence, and urethral diverticulum, with their easy surgical repair. The outcomes of single- and multistage repairs of proximal hypospadias are comparable; no technique can be considered better than any other. Thus, it is more judicious for a hypospadiologist to master a few of these procedures to achieve the best results, regardless of the technique used.

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* Corresponding author. Tel.: +20 122 3690597/5758575.

E-mail address: hithamalmewale@yahoo.com (H. Badawy).

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Introduction

The surgical reconstruction of distal penile hypospadias in a single stage is the standard practice for repairing anterior hypospadias. Unfortunately, it is not simple

to extrapolate the same principle to posterior hypospadias [1]. Numerous surgical techniques have been described for proximal hypospadias repair that can be broadly classified as single or multi-stage procedures.

The multi-stage technique was formerly adopted for its simplicity and safety more than for its effectiveness, but a single-stage repair is used by many surgeons and achieves a high success rate, being safe and effective [2]. Although accomplishable in a single stage, there are many complications, necessitating a second and sometimes third intervention.

Contemplating proximal hypospadias repair poses many challenges, as the release of ventral curvature and the simultaneous reconstruction of the urethra and skin represents an ongoing dilemma. No current evidence suggests the superiority of one surgical technique over another. There is still no consensus among hypospadiologists about whether a single- or multi-stage operation is the optimal treatment for proximal hypospadias. Is there a solution to this dilemma? In this review we try to present current reported data on single- and multi-stage repairs to find an answer to this difficult question.

Methods

We searched Medline, PubMed, Scopus and Ovid for relevant publications for the period 2000–2012, using the terms ‘proximal hypospadias’, ‘posterior hypospadias’ ‘single stage’, ‘multiple stage’ and ‘complications’ including the use of ‘all possible combinations’, and the use of ‘OR’ options. We included only series containing detailed results after a primary repair of proximal hypospadias. Articles not in English or duplicated publications and congress abstracts were also excluded.

Two doctors reviewed the abstracts. Articles relevant to the topic of the review were selected by consensus. Selected articles were distinguished according to level of evidence [3], and their quality was assessed [4].

The 86 articles retrieved from this search were analysed according to the technique of repair, success rate, number of included children, re-operative rate, and length of follow-up. Moreover, the selected articles were further analysed for the type of urethroplasty, penile straightening procedure, complications and outcome.

Discussion

Single-stage repair of proximal hypospadias

Achieving a repair in distal hypospadias was feasible with a very high success rate, exceeding 90% in most of the publications [5]. Achieving the same goals in a single stage in proximal hypospadias is not as certain as that for distal hypospadias [1]. Reports of the single-stage repair of posterior hypospadias in the last 10 years

showed many defects in these publications, including the retrospective nature, too few patients, a variable duration of follow-up, different methods of reporting the curvature, and moreover, the reporting of complications was not complete in most of the studies. The repair of proximal hypospadias in a single stage entails two important steps, i.e. the straightening of the penis (correction of ventral curvature) and the construction of the neourethra.

Correction of penile curvature

Authors adopting a single-stage repair try to preserve the urethral plate for either subsequent tubularisation, the incised-and-tubularised repair, or for the onlay of preputial flaps [6–8]. With the objective of preserving the urethral plate, penile curvature is managed using appropriate techniques in most of the cases.

Correcting penile curvature starts with the complete degloving of the penis, removal of any dysplastic dartos tissues, then inducing an artificial erection to assess the exact degree of curvature. When the curvature is severe, mobilisation of the urethral plate from the underlying corpus cavernosum is contemplated, followed by mobilisation of the normal proximal urethra up to the bulbar urethra, in what is described as radical bulbar dissection [9].

Bhat [10] reported the successful correction of penile curvature in a series after the above steps of urethral mobilisation. According to Bhat, the success rate was 89% (30 of 34 children) for correcting the curvature after urethral mobilisation. Bhat reported sufficient straightening of the penis in 74%, and in additional 15% he improved the straightening by glanular dissection of the plate. In that series Bhat resorted to transecting the urethral plate in only 6% and dorsal plication was needed in another 6% of the patients.

In the former series [10] the author resorted to transection of the urethral plate when the curvature remained severe and then used ventral corporoplasty. Instead of a ventral corporoplasty, Snodgrass and Prieto [11] reported on the use of multiple transverse incisions made in the ventral aspect of the corpus cavernosum at the point of maximum curvature, taking care not to damage the underlying erectile tissue, coupled in some of their cases with a small Nesbit dorsal plication to assure a completely straight penis. In their series, they had two groups of children. In group I, 45 children (early in the surgeons’ experience) had a complete transection of the urethral plate and had ventral penile lengthening in seven and transverse cuts in four to treat persistent severe curvature after the transection. In group II (23 patients) the urethral plate was preserved in all, coupled with multiple transverse corporal cuts. Hence the incidence of cutting of the urethral plate decreased significantly from 54% to only 15%, with no recurrence of curvature in a mean follow-up of 11 months.

However, despite every attempt to preserve the urethral plate, in some cases where the urethral plate is unhealthy, or severe penile curvature persists after the above procedures, we resort to transecting the urethral plate and performing a ventral corporoplasty, but according to Braga et al. [12] ventral grafting was only needed in 1.5% of their cases over 10 years, which is a similar experience to that reported by De Mattos e Silva et al. [13] in their series of 184 children, where only 18 needed a ventral corporoplasty to straighten the penis.

Ventral penile lengthening entails a transverse corporotomy from the 3 to 9 o'clock position, not involving the underlying cavernous erectile tissue. This incision leads to a diagonal defect after the penis is straight, which needs to be covered. Different methods of coverage were used, including grafts and flaps. Small intestinal submucosa, dermal graft, tunica vaginalis graft, buccal mucosa graft and others were used. Tunica vaginalis was tested as a flap, with good results [14–16].

We found no prospective study comparing the outcome of dorsal plication and ventral penile lengthening, but Braga et al. [17] reported a retrospective comparative trial between dorsal plication and ventral penile lengthening as the two major techniques for correcting penile curvature. They studied 100 children operated between 1996 and 2004, and classified them into two groups. Group I included 32 patients with ventral penile lengthening after urethral plate transection in 30, and group II included 68 with dorsal plication, 16 of them undergoing urethral plate transection only. Dorsal plication in the second group was performed as a Nesbit tuck in 42 and midline plication in 24. The rates of recurrent curvature at a mean follow-up of 2 years were significantly different between group II and group I, at 27.9% vs. 9.4%, respectively.

Different classification systems for curvature were used in the reports assessed in this review; in the above study [17], the classification of the American Academy of Pediatrics survey was followed, which classifies curvature into mild, moderate and severe, where mild curvature is $<30^\circ$, moderate $30\text{--}43^\circ$ and severe is $>43^\circ$, after complete degloving of the penis and removal of the dysplastic dartos tissues, and during an artificial erection [17].

Importantly for proper reporting and comparing different techniques addressing penile curvature, we recommend a unified approach for accurately defining the curvature, that should be followed and described in reports dealing with this pathology. Mingin and Baskin [18] recommended that after degloving the penis and dissecting the ventral aspect of the corpus cavernosum, if the curvature is $>30^\circ$ then the urethral plate should be mobilised. If the curvature decreases to $<30^\circ$, other authors suggested leaving the minimal curvature or using dorsal plication. If the curvature remains $>30^\circ$

they suggested transecting the plate, followed by proximal mobilisation. If the curvature is distal in the shaft of the penis, then dorsal plication is the treatment. If the penis is small after androgen stimulation, and there is no space for dorsal plication (which will further shorten the penis), then they suggest urethral plate transection and proximal mobilisation.

Single-stage urethral reconstruction

In posterior hypospadias, after straightening of the penis, there are three techniques for urethral reconstruction, i.e., using the preserved urethral plate, not using the urethral plate, and urethral plate division and reconstruction.

Using the urethral plate

The Thiersch–Duplay procedure describes the tubularisation of the urethral plate after two parallel longitudinal incisions around the edges of the plate. Amukele et al. [8] reported their experience with 265 consecutive cases of proximal penile hypospadias treated using the Thiersch–Duplay technique. The mean age at surgery was 7 months and the mean follow-up was 75 months. They reported a success rate of 88.7% and only an 11.3% complication rate, in the form of fistula, meatal stenosis, concealed penis, diverticulum and dehiscence. All complications were repaired in a second stage, with a perfect outcome. The incidence of fistula decreased from 17% to 1.8% in the last 126 children, after using a second-layer cover of tunica vaginalis in 39 and dartos in 125 children.

The tubularised incised-plate (TIP) urethroplasty, which is a modification of the Thiersch–Duplay technique, remains one of the most successful methods for posterior hypospadias. Snodgrass et al. [19–21] reported on 36 children, 13 with proximal penile hypospadias, 11 with penoscrotal, nine with scrotal and three with perineal hypospadias. Fourteen of the children received preoperative testosterone stimulation. They used a TIP repair in 26 and in two stages in 10 children. The authors reported a 13% complication rate, in the form of one case of strictured urethra and one glanular dehiscence, with an overall success rate of 87%. The follow-up was 12 months in 24 children. They showed an improvement in the results with accumulating experience in posterior hypospadias, by using a double-layer closure of the urethral plate and by using the tunica vaginalis as the second layer.

Ghanem and Nijman [6] reported on the TIP repair in 47 children with proximal penile hypospadias, with only a 12% complication rate, in the form of four with a fistula, one meatal stenosis and one glanular dehiscence, all of them repaired sufficiently in a second stage. Bhat et al. [22] reported on 27 children (mean age 6 years 4 months) having proximal hypospadias repaired using

a TIP and preputial reconstruction, with only two fistulae and preputial dehiscence in one case. Previous studies show that even in the severe form of hypospadias it is possible to preserve the urethral plate after penile straightening, and it can then be used for TIP. Previous objections to one-stage procedures were obviated by the TIP repair, as it is safe, simple and can provide cosmetic and functional results comparable with the two-stage repair.

However, to our knowledge, we found no study addressing grafting in the Snodgrass technique for posterior hypospadias, which we think needs to be evaluated in a future prospective trial vs. the ungrafted Snodgrass technique.

The transverse-island preputial flap onlay over an intact urethral plate has stood the test of time. Patel et al. [23] reported the long-term follow-up of posterior hypospadias repaired by either onlay or a tube operated by one surgeon from 1981 to 1992. Of 125 children treated, 30 could be contacted. An island tube was used in 14 children and an island onlay in 16. The overall complication rate was 16.6%, including two fistulae with the tube, distal stenosis in one tube repair, and distal stenosis in two onlay repairs. The mean follow-up was 14.2 years. The functional and cosmetic outcomes were deemed satisfactory by the parents, patients and surgeons.

A double-faced onlay, which was modified to leave a strip of vascularised skin to be used as a cover over the neourethra, and thus prevent overlapping of the suture line, had a good success rate in 15 children, with only two having minor complications (one fistula and one dorsal skin dehiscence) [24].

Braga et al. [25] reported a retrospective comparative study between the TIP and transverse island onlay methods (35 TIP vs. 39 onlays). The overall success rate was 60% and 43%, respectively, after a mean follow-up of 30 and 38 months, respectively. The rate of recurrence of curvature, of 5.7% vs. 12.5, was statistically significantly different. The difference in the pattern of urinary flow was in favour of the onlay, with no documented true stricture in the TIP group. Hadidi [26] reported on 107 children (mean age 25 months) repaired using a lateral vertical flap. In a mean follow-up of 72 months, the complication rate was 9%, in the form of three with a fistula, two with proximal stenosis, and four with a diverticulum.

Not using the urethral plate

In severe forms of posterior hypospadias necessitating transection of the urethral plate it is still possible to accomplish the repair in one stage. Rigamonti and Castagnatti [27] described the onlay of a preputial island flap directly on the corpus cavernosum in 14 children (mean age 16 months). Over a mean follow-up of 7 months the complication rate was only 21%, comprising one fistula, one diverticulum, and one partial dehiscence,

all repaired effectively in one minor stage. Aoki et al. [28] reported on a tubularised transverse island flap repair in 22 children (mean age 17.5 months) with a complication rate of only 13.6%, in the form of meatal stenosis in two and urethral fistula in one, over a mean follow-up of 18 months.

Macedo et al. [29] described the 'three-in-one' technique using buccal mucosa, an onlay with a vertical skin flap, and a tunica vaginalis cover, in 40 children. The complication rate was 37%, including four with meatal stenosis, four with a diverticulum, five with fistula, two with residual curvature and two with recurrent UTIs, with a re-operative rate of only 31.5% for a second surgery. Djordjevic et al. [30] reported a combined buccal mucosa and dorsal vertical skin flap in 17 patients, with minor complications in three, over a mean follow-up of 25 months. In the last two reports, the surgeon used the buccal mucosa as a substitute for the transected urethral plate, and made an onlay over it, with an excellent success rate.

Severe hypospadias, even in those associated with scrotal transposition and a bifid scrotum, can be repaired in one stage, with an acceptable complication rate. DeFoor and Wacksman [31] operated on 20 children with severe posterior hypospadias and penoscrotal transposition (median age 10 months), 17 of whom were treated using the HodgsonXX technique, and the remaining three were repaired using the Koyanagi technique (which uses paramental vascularised skin flaps to construct the neourethra after transecting the urethral plate). Over a mean follow-up of 23 months, the authors reported a 20% complication rate, comprising two of fistula and two of diverticulum, which are minor complications. Hayashi et al. [32] reported on 12 children treated with a modified neo-Koyanagi repair, with only one complication, a fistula. Nerli et al. [33] reported on 14 children who had a repair using the modified Koyanagi method, with five complications, i.e. one dehiscence, three with fistula and one meatal stenosis. Except for the dehiscence, the other complications were minor.

Reported trials comparing some of these techniques were retrospective. De Mattos et al. [34] retrospectively analysed 184 children operated by three techniques, with 133 repaired by onlay, 25 using buccal mucosa, and 26 with the Koyanagi method. Fortunately only 18 of them required ventral penile lengthening. The complication rates were 28.5%, 56% and 61.5%, respectively.

Urethral plate division and reconstruction

Vella et al. [35] described a one-stage onlay technique for urethroplasty in severe hypospadias with chordee. The urethral plate was divided obliquely into two flaps. The incision was extended into the penile skin to elongate both strips of transversally divided urethral plate. Chordee was excised and the penis was straightened. The elongated urethral plate strips were re-anasto-

mosed. An onlay flap was fashioned and the neourethra was constructed. This procedure was used in six patients. After the 10-month follow-up a fistula developed in only one patient, and all six patients had an excellent cosmetic result.

Hayashi et al. [36] reported on the onlay-tube onlay in six children, after division of the urethral plate. There were minor complications, comprising one fistula and one meatal stenosis.

Multi-stage repair of proximal hypospadias

The use of multi-stage operations for hypospadias repair remained the norm in many centres, despite the criticism of exposing the child to several operations. Recent reports suggest that this type of reconstruction achieves excellent functional and cosmetic outcomes [37].

In those cases where it is necessary to transect or excise the urethra or urethral plate, and thus create a full-circumference defect, the value of the two-stage repair is indisputable. Proponents of the staged repair suggest that using this technique can effectively correct chordee, can help to design the future plate for the second-stage urethroplasty, and hence in their view achieve the best cosmetic outcome of the glans and penis, even better than after the single-stage repair [2,38]. This proposal is based on the assurance that the penis is fully straight, that the graft will take, and that the tissues will become well vascularised in preparation for the urethroplasty at the second stage [2].

The multi-stage repair entails two important steps, as in the single stage repair, which are straightening the penis, and preparation of the ventral bed for the second stage neo-urethral reconstruction.

Straightening of the penis

The penis is degloved as in the single-stage repair, with removal of all dysplastic dartos tissues on the ventral aspect of the corpus cavernosum. The ventral dissection can be extended proximally to the bulbar region [39]. After that, some advocate corporeal rotation or dorsal plication to correct any residual curvature [9]. Others recommend extending the dissection underneath the urethral plate (urethral plate mobilisation) [9].

If the residual curvature is mild after ventral dissection, dorsal plication appears to be the most frequently used approach [39,40]. However, in cases with persistent severe curvature, most authors recommend sectioning the urethral plate. By comparison, if a curvature of $>30\text{--}40^\circ$ persists even after plate mobilisation/transection, ventral penile lengthening should probably be used, by grafting the corpus cavernosum defect, using one of many types of graft noted previously, with correction of the penile curvature as in the single-stage repair [18].

Preparation of the ventral bed for the second stage neourethral reconstruction

Two techniques are in current use, i.e. Byar's preputial flaps or a ventral graft over the corpus cavernosum.

For Byar's preputial flaps, after the urethral plate is transected, the dorsal prepuce is incised in the midline and brought ventrally to be sutured in the midline ventrally, extending from the proximal urethral meatus to the tip of the glans penis. After a variable period of 0.5–1 year, the second stage closure is made [41]. Gershbaum et al. [42] reported on 11 children with proximal hypospadias and a follow-up of 60–180 months. There were complications in only two children, i.e. one fistula and one urethral diverticulum.

Arshad [43] reported the largest series, including 100 children, in whom Byar's preputial flaps were used. Eighteen children had complications, i.e. a fistula. In this series the authors did not state the number of patients with a complete follow-up, or the duration of the follow-up.

Cheng et al. [44] transected the urethral plate proximally, preserving the distal portion, and used Byar's flaps to bridge the gap to the native urethra. The authors reported on 14 children followed for 6–36 months. Two children had complications, i.e. one with a fistula and diverticulum, and one with a diverticulum. Aseem et al. [45] operated on 10 children, with a mean follow-up of 41.5 months, and seven had complications, all with a fistula, three with meatal stenosis and one with a urethral diverticulum.

It is clear in these studies that the complication rate is very variable, at 14–70%. The work load of the surgeon is clearly related to this incidence, which is greater with a lower work load. Complications are not major if they are in the form of a fistula, meatal stenosis and diverticulum, and thus no major complications (e.g. complete dehiscence) were reported in these studies.

Ventral grafts over the corpus cavernosum are a valid option when the penis is totally straight, when the curvature is $<30^\circ$, or if several superficial transverse corporotomies are used for correcting the ventral curvature with no corporeal grafting. The glanular wings are made, the graft is laid open on the ventral aspect, and well secured to the bed to enhance neovascularisation. Grafts could be harvested from several sites, including the inner prepuce or buccal mucosa.

The inner prepuce is an ideal urethral substitute, as it is very thin and flexible, takes reliably, is designed to be moist, has no potential for hair growth and the donor site is both convenient and expendable. Ferro et al. [46] reported on 41 patients repaired in two stages, using preputial grafts in the first stage, with a follow-up of 1–46 months. Both stages of the repair were completed in 34 children. There were complications in eight patients (24%), comprising four with glans disruption, two with a coronal fistula, one urethral diverticulum and urethral stenosis due to balanitis xerotica obliterans in one.

There were no complete disruptions, and the functional and cosmetic outcomes were satisfactory over the follow-up. Johal et al. [47] reported on 62 boys who underwent a two-stage preputial graft repair (median age 27.6 months, median follow-up 26 months). There were no complications in the first stage, but after the second stage there were three boys each with glans dehiscence, residual curvature or meatal stenosis.

Oral mucosa, whether harvested from a buccal, labial or lingual donor site, is at present the most widely used alternative to the inner prepuce. Although it has a more variable graft take and behaviour during maturation, it remains a very good substitute for the urethra, producing no visible scarring or significant morbidity at the donor site, and obviously no hair growth. Buccal mucosa is bulky and must be thinned considerably, but large amounts of mucosa can be taken from the cheeks, usually enough for replacing the complete circumference of the penile urethra. The donor sites in the cheek can usually be closed directly to promote patient comfort.

Snodgrass and Elmore [48] reported on 25 children with a two-stage buccal mucosal graft who had undergone a previous unsuccessful hypospadias surgery. To the date of the publication, 20 children had undergone a second-stage closure with no meatal stenosis, stricture or diverticulum, and only a fistula in one and partial glans dehiscence in four. All children with complications had corrective surgery and finally had a cosmetically and functionally acceptable penis.

Long-term studies of functional outcomes are essentially available only in patients who have undergone a two-stage procedure, as this was the most common approach before the 1980s. In the study of Aulagane et al. [49] voiding abnormalities were the leading symptoms; 37% of patients had voiding difficulties, none had erectile problems, and only 11% had ejaculatory problems. The cosmetic appearance was satisfactory, as 70% had an apical meatus and 82% had a fully straight penis. Lam et al. [50] followed 25 patients repaired using Belt–Fuqua technique, and assessed them after puberty; of these, 10 had to ‘milk’ their urethra after voiding, and of the 20 able to ejaculate, nine had to ‘milk’ the urethra of ejaculate.

Conclusions

From this review of the repair of posterior hypospadias in the last 10 years, most studies are mainly retrospective, and incomplete in many of the details about the exact position of the meatus, the degree of curvature before and after degloving of the penis, full operative descriptions, postoperative immediate and delayed complications, and the follow-up.

Comparative studies between different techniques of single-stage repair are almost retrospective, not randomised, full of bias and with a very weak level of evidence.

There are no comparative studies with a prospective design between single- and multi-stage repairs. Long-term follow-up studies are very scarce, with most of them being retrospective and with many patients lost to follow-up.

It is clear that the complication rate of a single-stage repair of proximal penile hypospadias is 8–61.5%. This wide variation depends on the surgeon’s skill, workload and experience. In the multistage repair we found that the incidence of complications was 15–70%. The re-operative rate was also similar for the two procedures, but in the multi-stage procedure the child is exposed to more surgery than in the single-stage operation, with the same incidence of complications and re-operative rate.

Both techniques showed a similar pattern of post-operative complications, in the form of fistula, meatal stenosis, partial glans dehiscence, and urethral diverticulum. Fortunately, the serious result of having a complete dehiscence of the repair, even in the most severe forms of posterior hypospadias, using either the single- or the multi-stage procedure, is very uncommon.

As with any review, the present study has limitations. We did not account for some variables such as any associated morbidity and its influence on the success of surgery, the use of additional coverage for the urethroplasty, and the type of drainage and care before and after surgery. These factors might influence the surgical outcomes and have significantly changed during the last few decades. However, such confounding factors are not often readily accessible, due to the ambiguity in reporting by authors. More importantly, the cost-effectiveness of both techniques was not discussed in any of the reports, stressing the need to address this issue in future studies.

Hypospadiologists should be aware of the evidence supporting any single intervention, to standardise their management policies. The careful selection of patients is mandatory to avoid unnecessary operations for those who might benefit from simpler and equally effective procedures, not only in terms of clinical outcome, but also of hospital stay and costs.

The outcomes of single- and multi-stage repairs of proximal hypospadias are comparable; no technique can be considered better than another. Thus, it is more judicious for a hypospadiologist to master certain procedures and accumulate excellent experience in few of these techniques to achieve the best results, regardless of the technique used.

Conflict of interest

None.

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None.

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