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Chordee Without Hypospadias: Report of 79 Chinese Prepubertal Patients

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Abstract

A series of Chinese prepubertal patients with congenital chordee without hypospdias is presented and the clinical data described. From July 1999 to September 2006, 79 boys with congenital chordee without hypospadias were treated in the Department of Pediatric Surgery, West China Hospital of Sichuan University, China. The ages ranged from 21 months to 14 years, with a mean of 76.8 months (6.4 years). The patients were

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categorized according to structural defect into 4 groups, with the aid of intraoperative artificial erection. Group I included those with skin tethering (28 cases, 35.4%); group II, fascial chordee (22, 27.8%); group III, corporal disproportion (10, 12.7%); and group IV, urethral tethering (19, 24.1%). Chordee-related structural defect was considered the only criterion for classification, and urethral dysgenesis influenced the choice of surgical procedure. The chordee in group I patients was corrected with penile degloving; group II, release of dense fibrous tissue in addition; group III, dorsal-midline-plication-based correction; and group IV, longitudinal-island-flap-urethroplasty-based repair. At a mean follow-up of 14.8 months (range, 2 to 63), all patients had penile straightening except 1 group III patient with residual curvature that was managed upon reoperation. Glans dehiscence occurred in 1 group II patient who underwent a tubularized incised plate urethroplasty. Urethrocutaneous fistula and urethral stricture were found in 2 group IV patients who underwent island flap urethroplasty. With the categorization based on structural

defect, chordee without hypospadias may be managed well with minimized complications.

Key words: Surgery, penis, orthoplasty

Congenital chordee or ventral penile curvature is common in hypospadias, whereas it is much less common when the meatus is orthotopic. Since the understanding of hypospadias has developed, it has become well acknowleged that urethral tethering is not always the cause of penile curvature, and this has led to more urethra-preserving urethroplasty procedures. The same is true for isolated chordee. Since we agreed with the theory that chordee should be corrected according to anatomic changes, these principles have been adopted in our clinical practice in management of chordee with and without hypospadias. With such background, a series of Chinese prepubertal patients with chordee without hypospadias is presented here and the clinical data described.

Materials and Methods

Patients

From July 1999 to September 2006, 79 boys were treated for chordee without hypospadias in this institution. The age at operation ranged from 21 months to 14 years, with a mean of 76.8 months (6.4 years). Patients with previous surgical corrections were excluded. Indications for surgery included parental

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complaint of penile curvature at erection and/or a penile angle greater than 30 degrees on examination under natural erection.

Surgical Technique

A circumcision was made, and the skin was degloved down to the base of the penis. Then an artificial erection was induced with injection of normal saline into the corpora cavernosa to identify the presence of residual curvature. Patients whose penis straightened after degloving were considered to have group I chordee (skin tethering). When chordee persisted, any abnormal dense fibrous tissue was excised over the urethra. If release of fibrous tissue resulted in satisfactory orthoplasty, group II chordee (fascial chordee) was designated. For patients of groups I and II, penoplasty was performed after orthoplasty. For patients in whom the 2 steps failed and the curvature was not thoroughly resolved, the urethra and corpora cavernosa were checked for the cause of curvature. If the penis was still ventrally curved under artificial erection, although no significant urethral tethering was shown, the cause was ascribed to a disproportion of dorsal and ventral aspects of the corpora cavernosa (group III chordee, corporal disproportion), and the patients were treated with dorsal midline plication (DMP). On the contrary, when the urethra was found tethering the corpora cavernosa under artificial erection and forming a bow to string configuration, group IV chordee (urethral tethering) was considered and urethral replacement warranted. For group I, II, and III patients, if the distal urethra was dysgenic and showed a lack of corpus spongiosum, a tubularized incised plate (TIP) urethroplasty was done. For group III patients, when dorsal tunica albuginea plication was not enough to straighten the penis and/or DMP alone for curvature correction might lead to significant penile shortening, additional urethroplasty was done. Similarly, for group IV patients with severe curvature that was not likely to be corrected through a single urethroplasty, a DMP procedure was completed in addition. At the end of surgery, an artificial erection was done to confirm penile straightening.

Postoperative dressing and stenting varied according to surgical procedure. For patients who did not

undergo urethroplasty, the urethral stent was removed 3 days after surgery and the dressing was changed and finally removed at 7 days. For those who did undergo urethroplasty, the urethral stent was reserved for 9 days. The dressing was changed at 3 days and removed at 9 days.

On follow-up, the correction of penile curvature was evaluated by physical examination and patient and/or parental observation of erection.

Results

Twenty-eight patients (35.4%) were categorized as group I chordee, 22 (27.8%) as group II, 10 (12.7%) as group III, and 19 (24.1%) as group IV $(\underline{\text{Table 1}})$. Ventral skin was deficient in all the patients except 2 group II patients and 1 group I patient with intact prepuce. Some associated anomalies were noted, including webbed penis, penile torsion, penoscrotal transposition,

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undescended testis and hydrocele. These were described with corresponding chordee groups as listed in <u>Table 2</u>. Such anomalies were treated simultaneously, and no related complications were found at follow-up.

View this table: Table 1. Clinical data of patients

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View this table: Table 2. Associated anomalies

Follow-up was 2 to 63 months (mean, 14.8 months). Of the 79 cases, 75 (94.9%) were cured after 1 operation. All group I patients were successfully treated with 1 operation. In 1 case of group II patients who underwent a TIP urethroplasty, glans dehiscence developed 10 days after the operation and a repeat TIP urethroplasty 8 months later was successful. Chordee recurrence was found in 1 group III patient who had a 90-degree curvature and underwent DMP plus longitudinal island flap (LIF) urethroplasty; this patient was managed with another DMP operation 1 year later. Complications occurred in 2 group IV patients, including urethrocutaneous fistula in 1 patient who underwent LIF urethroplasty and urethral stricture in 1 who underwent LIF plus Duplay procedure. The 2 patients were treated successfully with a second repair. No significant penile shortening was reported.

Discussion

The precise etiology of congenital chordee has not been elucidated to date, although it is now well accepted that varied anatomical changes are related

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to chordee with and without hypospadias, and various surgical procedures are required. Young (1937) advocated that chordee was due to a congenitally short urethra and that the urethra should be transected. Years later Devine and Horton (1973) raised their theory that chordee without hypospadias resulted from dysgenesis of

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fascia surrounding the urethra. They further classified chordee into 3 groups according to the layers affected, namely group I (with deficient corpus spongiosum, dartos, and Buck fascia), group II (with deficient dartos and Buck fascia), and group III (with only dartos fascia affected). Kramer et al (1982) found that corporal disproportion was another principal cause of chordee, and they classified this condition as group IV chordee without hypospadias. This classification system had been widely adopted until 1998, when Donnahoo et al (1998) presented their series of 87 patients with penile curvature without hypospadias. They proposed a systmetic approach to the surgical management of chordee without hypospadias and divided this anomaly into 4 groups according to structural defect and surgical steps. Their treatment algorithm helps to define the cause of isolated chordee in a stepwise fashion and thus minimizes the need for risky urethral replacement. The categorization system recommended by Donnahoo et al (1998) was adopted in our institution with

some modifications. When the urethra was abnormal, they believed that 2 entities should be treated—

either a congenital short urethra with dense fibrous tissue beneath or a hypoplastic distal urethra lacking the corpus spongiosum. The former was included as group IV isolated chordee and was treated with division of the urethra and creation of an interposition graft. The latter was excluded because such patients were thought to have poor-quality distal urethras, which required excision of the abnormal urethral segment and subsequent reconstruction. According to the categorization described by Kramer et al (1982) and formerly Devine et al (1973), the latter condition was ascribed to urethral dysgenic tethering and the treatments of choice included division of the urethra with urethral reconstruction or mobilization of the anterior urethra with excision of the underlying fibrous tissue (Kramer et al, 1982) or alternatively preservation of all the layers around the local dysgenic distal urethra (Hernandez et al., 2001). All congenital chordee patients, including those with chordee with orthotopic meatus, should be included in the classifications (ie, a single criterion for classification should be insisted upon), and chordee and urethral abnnormality should be corrected as 2 relatively isolated problems, similar to hypospadias correction (Baskin and Ebbers, 2006). When the distal urethra is dysgenic and contributes little to penile curvature, a urethra-preserving urethroplasty is a rational choice; when an abnormal urethra tethers the corpora cavernosa, urethral replacement is warranted. In our series, a hypoplastic distal urethra segment was noted in 1 group I patient, 3 group II patients, and 1 group III patient. However, the hypoplastic segment was not considered responsible for the chordee; thus, the dorsal healthy urethra was preserved and a TIP procedure performed. Urethral replacement is necessary for group IV patients. Various options are available, and the technique selection is influenced by several factors and dependent on the surgeon's experience and preference (Cook et al, 2005). In our clinical practice, the principal technique for urethral replacement is based on the LIF procedure (Chen et al, 1993; Duckett, 1998). The success rate for 1 operation being sufficient to fix the chordee was 89.5% in this series, which is comparable to that reported by other authors (Scuderi et al, 2006;

disproportion (Bologna et al, 1999). The Nesbit (1965) technique was formerly a popular method for the correction of penile curvature. Based on their own anatomic and embryologic investigations, Baskin et al (2000) developed a DMP technique. According to their findings, the dorsal midline area is nearly nerve free and the tunica albuginea is thick. The DMP procedure is theoretically and practically acceptable and has been widely applied (Bar Yosef et al, 2004; Soygur et al, 2004; Yucel et al, 2006). In the current series, all of the patients (n = 12) who underwent a DMP had a good

Great variety exists as to the dorsal correction of chordee or the treatment for corporal

Duckett, 2002).

outcome except 1 group III patient with recurring curvature that was managed with another DMP. In our experience of correction of chordee with and without hypospadias, multiple plication might be effective for postpubertal patients; however, for prepubertal patients with undeveloped penises, DMP remains the mainstay of dorsal correction. This series represents no experience at this institution with more complicated procedures, such as corporal rotation and penile disassembly (Dectar, 1999; Dessanti et al, 2002; Perovic et al, 1998).

To date, none of our patients were followed up to the postpubertal stage; therefore, data on erectile function and related symptoms were limited. Long-term follow-up is still needed for outcome evaluation.

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