Epidemiology of Male Genital Abnormalities: A Population Study

WHAT’S KNOWN ON THIS SUBJECT: There are misconceptions regarding childhood phimosis. Textbooks still teach that male children should have retractable foreskin by age 3. Young children are referred for evaluation for phimosis, which is a commonly used diagnosis for postneonatal circumcision.

WHAT THIS STUDY ADDS: We found a high prevalence of physiologic phimosis in kindergarten children, up to 44% at age 6. We also reviewed the incidence of other congenital abnormalities in this coastal Chinese city. The management and complications of these conditions were analyzed.

abstract

BACKGROUND: We conducted genital health wellness screens in male kindergarten children between the ages of 3 and 6 years to assess the incidence of congenital abnormalities and their treatment.

METHODS: We performed genital examinations on 2241 male children in 8 kindergartens. We screened for 4 conditions: phimosis, hypospadias, cryptorchidism, and hydrocele/hernia. We assessed the incidence of these conditions and the effectiveness or lack of their treatment.

RESULTS: Among this sample, 55.5% children aged 3 to 4 years and 44.1% aged 5 to 6 years were found to have persistent phimosis. The circumcision rate, excluding those performed in conjunction with hypospadias repair, was 2.8%, but it carried a 3.2% complication rate. There was a lower incidence of hypospadias and cryptorchidism than reported in the literature at 0.2% and 0.4%, respectively. Our hypospadias repair rate was 60%, with a success rate of 66.7%. Our cryptorchidism repair rate was only 25%, and all repairs were performed above the age of 5 years. Incidence of hydrocele and hernias was 1.2%, and our treatment rate was 46.2%. Finally, we found high incidence of keloid formation, 73.3%, associated with inguinal incision.

CONCLUSIONS: There was high prevalence of phimosis in Chinese boys, a natural physiologic condition, up to age 6. There appeared to be lower incidences of hypospadias and cryptorchidism in our screened population. However, there were opportunities for us to improve the diagnosis and treatment of these 2 conditions. Our hydrocele/hernia incidence was on par with literature, but we had a lower treatment rate. Finally, we found a high incidence of keloid formation associated with inguinal incision. Pediatrics 2014;133:e624–e627

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KEY WORDS
phimosis, hypospadias, cryptorchidism, hydrocele, hernia, keloid

Dr Wan designed the study and performed all examinations on the children; Dr Wang assisted in the study design, performed the administrative and coordinative duties for the study, and reviewed the results; and Dr Gu reviewed and approved the study and findings and served as liaison and reported findings to the local health department.

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In spring 2013, the Health Department of the District of Xiaoshan in the city of Hangzhou, Zhejiang, China, launched an initiative for male genital health wellness screen in kindergarten children to assess the incidence of congenital genital abnormalities and to evaluate the effectiveness of the local health facilities in the management of these conditions. Xiaoshan District has a population of ~2 million. It has 3 tertiary hospitals, 4 secondary hospitals, and ~50 rural community clinics, which have limited inpatient and surgery capabilities. The authors volunteered to perform the screening for this worthwhile program. We chose to screen for 4 congenital conditions: phimosis, hypospadias, cryptorchidism, and hydrocele/hernia. Because neonatal circumcision is almost never performed in China, we had the unique opportunity to observe the natural history of phimosis. We elected to use a strict definition for the phimosis; namely, the prepuce was considered to be phimotic only if it could not be retracted beyond exposing the urethral meatus (type I and II per the Kayaba classification). This finding would be considered pathologic in a nonjuvenile population. A prepuce that could be retracted beyond the urethral meatus with or without any physiologic adhesion was not considered to be phimotic (Kayaba classification type III–V). We classified the hypospadias into 4 categories: distal (glanular/coronal), midshaft, penoscrotal, and perineal hypospadias. In cryptorchidism, the retractile testis was excluded. For hydrocele/hernia, we included both communicating and noncommunicating varieties. We considered all hydroceles and hernias congenital.

METHODS

We screened 2241 children between the ages of 3 and 6 years in 8 kindergartens with permission from children’s parents and the school administrators. Fifty-one children did not participate in the screen and were not included, 45 for absenteeism and 6 for refusing the examination. A written report of any abnormal finding was given to the parents, and appropriate referral was made for the treatment and/or follow-up. In addition, the findings were reported to our health department. We divided the children into 2 groups, age 3 to 4 (range 2.5–4.5) years and 5 to 6 (range 4.5–6.5) years. We had 100% participation with the exception of the 45 absentees and 6 children who refused examination despite gentle coercion from the teachers. Five of the 6 children that refused the examination were from the 3- to 4-year-old group. All the examinations were performed by the senior author (Wan) to ensure consistent findings. The examination was performed in a designated classroom with the child standing. However, if there was any question in the case of phimosis or hydrocele/hernia, the examination was repeated in supine position. All children with nonpalpable testis in the scrotum were reexamined in a supine and frog-legged position to reduce the cremasteric reflexes. If the child was extremely agitated, we usually let him to lie in the bed for 10 to 15 minutes to calm down while observing his fellow classmates being examined. A testis that could be brought into and remained in the scrotum was considered a retractile testis.

RESULTS

The results are summarized in Table 1. In the 3- to 4-year-old group, we identified 622 children with phimosis and 497 without phimosis, 55.5% and 44.5%, respectively. There were 466 children with phimosis and 590 without phimosis in the 5- to 6-year-old group, 44.1% and 55.9%, respectively. Sixty-two children had undergone circumcision, excluding those performed in conjunction with hypospadias repair; a rate of 2.8%. We found 2 children with moderate to severe iatrogenic phimosis after circumcision. One child with midshaft hypospadias underwent circumcision, an inappropriate treatment. One child had significant residual adhesion along the coronal sulcus. The total complication rate for circumcision was 3.2% (2 of 62, excluding the coronal adhesion and the circumcision done for hypospadias). We identified 5 children with hypospadias, 3 distal and 2 midshaft, with a calculated incidence of 0.2%. Among the 3 distal hypospadias, 1 had successful repair, 1 had a failed repair, and 1 remained untreated. Of the 2 midshaft hypospadias, 1 had successful repair and 1 had circumcision. The repair rate for hypospadias in our community was 60% (3 of 5), and the success rate was 66.7% (2 of 3). We also noted 4 children with mega-urethral meatus with intact prepuce. However, because of the high prevalence of phimosis, the true incidence of this condition could not be assessed. We identified 9 cases of cryptorchidism in 8 children with 1 being bilateral. The incidence for cryptorchidism was 0.4%. Two children in the 5- to 6-year-old group with unilateral cryptorchidism underwent orchidopexy. One was successful; the other had severe testicular atrophy. None of the 3- to 4-year-old children had orchidopexy, for a total repair rate of 25%. There were 27 cases of hydrocele/hernia noted in 26 children with 1 being bilateral. The incidence for hydrocele/hernia noted in 26 children with 1 being bilateral. The incidence for this condition was 1.2%. Two of the hydrocele/hernia cases occurred in the children who had contralateral repair. There were 13 hydrocele/hernia repairs in 12 children with 1 being bilateral, a repair rate of 46.2%. We also noted 1 child who had undergone scrotal orchietomy for unknown reason; 1 child had surgery for torsion of the testicular appendix; and 1 child was found with a scrotal mass, probably a mass at the tail of the...
TABLE 1 Summary of Findings

<table>
<thead>
<tr>
<th>Category</th>
<th>Total no. children screened</th>
<th>Phimosis</th>
<th>No phimosis</th>
<th>Circumcision</th>
<th>Hypospadias</th>
<th>Distal</th>
<th>Midshaft</th>
<th>Repaired</th>
<th>Cryptorchidism</th>
<th>Repaired</th>
<th>Hydrocele/hernia</th>
<th>Repaired</th>
<th>Keloid formation</th>
<th>Total no. children not included</th>
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<td></td>
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<td>1151</td>
<td>622/1110</td>
<td>498/1110</td>
<td>29/1049</td>
<td>3/2</td>
<td>3/6</td>
<td>2/1</td>
<td>3/6</td>
<td>3/6</td>
<td>8/19</td>
<td>3/10</td>
<td>2/3</td>
<td>16</td>
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<td></td>
<td>466/1056</td>
<td>590/1056</td>
<td>33/1089</td>
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<td>1080</td>
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DISCUSSION

Children are born with phimosis, a natural physiologic condition. Throughout childhood, most if not all phimosis will resolve spontaneously. Oster’s study on Danish schoolboys showed a 1% incidence of persistent physiologic phimosis in 16- to 17-year-old adolescent boys. The glans penis and the prepuce are naturally adhered to each other at birth. During normal development, the glans penis and the prepuce will separate. The accumulation of the desquamated epithelial cells and sebum from this separation collects between the glans penis and prepuce, forming smegma. The natural separation of prepuce from glans penis, differential growth between penile shaft and the prepuce, and normal erections of the penis result in gradual enlargement of the preputial orifice, and hence the resolution of phimosis. The rate of this resolution varies from individual to individual, and the process will continue until the pubertal development is completed. Circumcision is one of the most commonly performed genital operations in the United States, and phimosis is the most commonly used diagnosis for post-neonatal circumcision. This was also true for our cohorts in whom 62 circumcisions were performed, primarily for “phimosis.” There exist conflicting data on the development and natural history of the foreskin. Furthermore, there is also controversy regarding pediatric circumcision, with the most vocal opponents against circumcision coming from Australia and the United States. No guidelines have been developed for circumcision. In 2012, the American Academy of Pediatrics issued a policy statement regarding neonatal circumcision: “Evaluation of current evidence indicates that the health benefits of newborn male circumcision outweigh the risks and that the procedure’s benefits justify access to this procedure for families who choose it.”

Furthermore, the 1999 Task Force on Circumcision reported that in 90% of uncircumcised males, the foreskin is retractable by age 5 years. This statement was based mostly on Oster and Gairdner’s publications and differs significantly from our current finding. The Task Force also noted a lack of data on complications of circumcision. According to our data, the incidence of iatrogenic phimosis after circumcision was 3.2%, higher than the incidence of pathologic phimosis in boys who were not circumcised by age 15 years. Of the 2 children who had moderate to severe iatrogenic phimosis after circumcision, the causes were likely due to the inadequate amount of foreskin that was removed and cicatricial scarring. Both children will probably require revision of the procedure. In the child with midshaft hypospadias who underwent circumcision, the hypospadias may not have been recognized, which would probably cause significant morbidity in the future. The overall circumcision complication rate in our study would be even higher at 4.8% if the circumcision done for the hypospadias was considered a complication. In 1949, British pediatrician Douglas Gairdner reported that “80 percent of boys should have retractable foreskin by age of two years old and ninety percent by age three.” This conclusion has been quoted in many textbooks and medical literature even today, including the latest edition of Campbell’s Urology: “Preputial retractability increases with age with 90% of uncircumcised boys 3 years of age with completely retractable prepuce and less than 1% by 17 years of age.” The International Classification of Diseases, Ninth Revision, has only 1 code, 605, for phimosis and no differentiation of the normal physiologic condition in children versus the pathologic condition found in adults, or phimosis resulting from inflammation, iatrogenic causes, or disease. North Carolina Medicaid classifies phimosis in children under age 6 years as normal. Even using this more strict definition, 44% of the children in our study would potentially meet the criteria of having abnormal foreskin. We believe the finding of nonretractable foreskin is not in itself a pathologic condition until pubertal development is complete. Therefore, it is our opinion that, at the least in Chinese children, treatment of phimosis in asymptomatic boys before age 6 is probably not warranted. It would be beneficial if more diagnostic codes were assigned to differentiate physiologic from pathologic phimosis. It is also important to remember that postneonatal circumcision may not be as innocuous...
a procedure as perceived, and general anesthesia is often required. There appear to be lower incidences of hypospadias\textsuperscript{10,11} and cryptorchidism\textsuperscript{12} in our study population than that reported in the world literature. We postulate that this may be correlated to the lower usage of estrogen/progesterone-based contraceptives in China.\textsuperscript{13} There appears to be opportunities for us to improve the management of hypospadias and cryptorchidism, especially the delay in the diagnosis and treatment of cryptorchidism. Our incidence of hydrocele/hernia was on par with the literature,\textsuperscript{14} but we seem to have a lower treatment rate. We found a higher than expected incidence of keloid formation associated with inguinal incision. Beside racial predilection for keloid,\textsuperscript{15} we posited that the surgical techniques used might also contribute to the high incidence of keloid formation.

**Conclusions**

On the basis of our screening data, 44% of the children at age 6 still had phimosis. This is a natural physiologic condition. Therefore, any intervention to “treat” this condition in asymptomatic boys before age 6 years is probably unwarranted. It would be beneficial if new diagnostic codes were assigned to differentiate physiologic from pathologic phimosis. It is important to remember that postneonatal circumcision might not be as innocuous a procedure as perceived, and general anesthesia is not be as innocuous a procedure as perceived, and general anesthesia is often required. There appeared to be lower incidences of hypospadias and cryptorchidism in our study population. We postulate that the lower incidence might be related to the lower use of estrogen/progesterone-based contraceptives in the Chinese population. We did find some deficiencies in our local pediatric urology care: (1) circumcision performed in our district carried higher than expected complications. (2) Although we did seem to have lower incidence of hypospadias and cryptorchidism, there were opportunities for improvement in their management, especially the delay in the diagnosis and treatment of cryptorchidism. (3) There was a high incidence of keloid formation associated with inguinal incision in our children. It may be advisable to review the surgical techniques used. This information and these recommendations were forwarded to our health department for review and tracking. We are planning to expand our male children genital health wellness screen into elementary schools and hope to gain a better understanding of the normal development of the prepuce as well as other congenital male genital abnormalities.

**References**

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