Research Article

Cryptorchidism in Infant & Children at Dr. Hasan Sadikin General Hospital Bandung

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ABSTRACT

Introduction: Cryptorchidism is a congenital disorder that occurs when one or both testicles do not descend completely toward the bottom of the scrotum. The incidence of cryptorchidism in Indonesia is 2.2% in term infants and 37% in preterm infants. Late management will have the effect of various unwanted complications such as hernias, testicular malignancy and torsion on the testicles, even infertility. The purpose of this study was to describe the clinical manifestation of cryptorchidism cases in infants and children at dr. Hasan Sadikin General Hospital (RSHS) Bandung for the period of 2015-2019.

Methods: This is a retrospective study from medical records in pediatric endocrinology clinic patient age 0-18 years who were diagnosed with cryptorchidism in RSHS during the period 1 January 2015 - 31 August 2019.

Result: A total of 125 patients with 91 medical records fulfilling the inclusion requirements and 31 excluded. From 91 cryptorchidism cases: 27 were bilateral, 36 were right unilateral and 28 were left unilateral. Hypospadias is the most common comorbidity, followed by retractile testes, hernia, hydrocele, phimosis and micropenis. The percentage of successful orchiopexy procedures performed at RSHS in this study was 100%.

Conclusion: The orchiopexy procedure performed in cryptorchidism at RSHS was 100% success rate. Patients with cryptorchidism mostly had late diagnosed at age more than 12 months old. The most common case is right unilateral cryptorchidism. Late diagnosis and late orchiopexy can have long term implications for fertility and growth of the testes. Therefore, ideally the procedure needs to be carried out before 12 months of age, and by 18 months at the latest.

Keywords: Cryptorchidism, Pediatric, Endocrinology, Testes, Comorbidities, Orchiopexy

INTRODUCTION

Cryptorchidism or Undescended Testis is a congenital genital disorder that often occurs in boys.[1, 2] Cryptorchidism occurs when one or both testicles do not descend completely toward the bottom of the scrotum. [1, 2] This condition can occur as a congenital abnormality or as an acquired condition which occur when the testes are located in the scrotum at the time of birth but migrate as the child grows.[1-3] Worldwide incidence rate in term infants is around 2-5% and 30% in preterm infants.[2–4] The incidence rate of cryptorchidism in Manado, Indonesia is 2.2% in term infants and 37% in preterm infants.[5] Study from Lubis SM and Pateda V, et al. revealed registries from North Sumatera, North Sulawesi, Jakarta and Bali provinces showed 29,6% boys under 6 moths old were diagnosed with cryptorchidism, the rest were diagnosed older.[6]

Late management of cryptorchidism will result in various complications such as hernias, testicular malignancy, testicular torsion, and infertility.[1-4] Maintaining the temperature of the testes for around 2-7°C below the body temperature is vital for spermatogenesis.[2] Increases in testicular temperature have an adverse effect to the production process of spermatozoa and can lead to infertility.[7] Testes that fails to descend into the scrotum until the 6th month are likely to remain in position and management is needed in the form of orchiopexy or administration of hormones to reduce the risk of complications.[9]

Success of orchiopexy procedure is defined when testis in scrotal position and lack of atrophy in testis. It depends on the type of cryptorchidism, either non-palpable or palpable, and the time of its surgery. The result of this treatment should be evaluated at least 1 year postoperatively. The most common complication of orchiopexy is testicular atrophy, this can occur due to truncated testicular blood vessels and inflammation of the surrounding tissue resulting ischemia and atrophy.[4,7,8]

Studies on the incidence and clinical manifestations of cryptorchidism in infants and children in Indonesia rarely been found. Literature study at RSHS Bandung on this topic has never been done. This study aims to describe the outcome of cryptorchidism cases in infants and children in RSHS from 2015 to 2019. Results from this study are also expected to be used for further research and scientific development.

METHODS

This study was conducted using a cross-sectional descriptive method in the Endocrinology clinic, Department of Child Health, RSHS Bandung Indonesia from 2015 to 2019. Samples were determined by total sampling. The data used were secondary data obtained from patient's medical record at RSHS Medical Record Center. The inclusion criteria were all infants and children with undescended testes with complete medical records that included data on age, type of cryptorchidism, testicular location, accompanying disorders, and management such as surgical and hormonal therapy from 1 January 2015 to 31 August 2019. Exclusion criteria were incomplete or missing medical record data, and medical records that are lost to follow-up. This study was approved by the Ethics Committee of Universitas Padjadjaran with registry number of 918/UN6.KEP/EC/2019 and received permission from Dr. Hasan Sadikin Bandung General Hospital with registry number of LB.02.01/x.2.2.1/13906/2019.

Data collection was initiated by selecting the medical records of patients diagnosed with undescended testis (cryptorchidism) and seeking treatment at the Endocrinology Clinic, Department of Child Health RSUP Dr. Hasan Sadikin Bandung. Data analysis was performed using Microsoft Excel, and then presented into results including the distribution of patient age and type of cryptorchidism, based on physical examination, distribution of locations, comorbidity, and surgical therapy.

The patient's age is defined as the age when diagnosed. The type of cryptorchidism is divided into unilateral and bilateral types. Right cryptorchidism (right unilateral) is defined by the presence of undescended right testis. Left cryptorchidism (left unilateral) is defined by the presence of undescended left testis. Bilateral cryptorchidism is defined by both undescended left and right testes. The location of cryptorchidism is inquired through history taking, physical examination, and through further examination such as ultrasound examination or laparoscopy exploration to find out the exact location of the testes which will then be classified into pre-scrotal, inguinal, intra-abdominal, or ectopic cryptorchidism.

Ectopic testes are those that descend outside the normal developmental path, causing them to be out of place. Retractile testes are those that have descended to the scrotum but are pulled back up by the strong cremaster muscle reflexes. Isolated cryptorchidism occurs when one or both testicles do not descend completely toward the bottom of the scrotum without any accompanying disorders. Accompanying disorders in this study are limited to the disorders that are associated with urogenital system and accompany cryptorchidism. Surgical therapy is performed with orchiopexy technique.

RESULTS

Based on the retrospective study conducted from 125 patients treated for cryptorchidism in the Endocrinology Clinic, Department of Child Health, RSHS Bandung, this study found 91 medical records that met the inclusion criteria and excluded 31 medical records that met the exclusion criteria, consisting of 16 missing or incomplete data and 15 medical records of patients who were lost to follow-up.

A total of 91 patients had cryptorchidism, with 85 patients had isolated cryptorchidism and 8 of them were accompanied by a retractile testis. Eighty-eight patients (96.7%) were diagnosed after they were 1 year old, which

were considered a late diagnosis. There were 3 (3.3%) patients diagnosed between the ages of 6 months to twelve months. Twenty-seven (29.7%) patients had bilateral cryptorchidism, 36 (39.6%) had right unilateral cryptorchidism, and 28 (30.8%) had left unilateral cryptorchidism from a total of 91 cryptorchidism patients. Of the 91 cases, only 52 (57.1%) cases were classified as palpable cryptorchidism through physical examination and 39 patients were nonpalpable. The age distribution of patients and physical examination results based on the type of cryptorchidism are listed in table 1.

No.	Variable	Cryptorchidism Type			Total	Percentage
		Left Unilateral (n = 28)	Right Unilateral (n = 36)	Bilateral (n = 27)		
1	Age (months)					
	≤6	0	0	0	0	0%
	6-12	1	1	1	3	3.3%
	≥12	27	35	26	88	96.7%
2	Physical Examination Results					
	Palpable	16	23	13	52	57.1%
	Nonpalpable	12	13	14	39	42.9%

Table 1. Overview of Patient Age and Cryptorchidism Type

Cryptorchidism cases in RSUP Dr. Hasan Sadikin Bandung were confirmed by supporting examinations such as ultrasound examination or testicular exploration using laparoscopy after a palpation examination. The distribution of cryptorchid sites varies from intra-abdominal, inguinal and pre-scrotal canals. Most cryptorchidism was in the inguinal canal with 45 cases occurring unilaterally. Due to the possible difference in the location of testes in bilateral cases, 19 cases of cryptorchidism of left testis and 18 cases of cryptorchidism of right testis occurred bilaterally in the inguinal canal. There were 23 cases of cryptorchidism that were nonpalpable. After confirming through testicular exploration by laparoscopy, the location of the testes was found to be intra-abdominal. The distribution of cryptorchid sites based on the supporting examinations is shown in table 1

Locations	Bilateral (n = 27)		Left Unilateral (n = 28)	Right Unilateral (n = 36)
	Left Testis	Right Testis		
Intra-abdominal	6	8	7	9
Inguinal Canal	19	18	20	25
Pre-scrotal	2	1	1	2
Total	27	27	28	36

Table 2. Distribution of Cryptorchidism Locations Based on Ultrasound or Laparoscopy

Table 3 illustrates the distribution of cryptorchidism based on comorbidities. Isolated cryptorchidism was found in 35 (38.5%) out of 91 cases of cryptorchidism. Hypospadias (17.6%) were the most common comorbidity, followed by retractile testis, hernia, hydrocele, phimosis and micropenis which became the most accompanying comorbidities in cryptorchidism at RSHS.

Ninety-one patients from this study had orchiopexy to return the testes to their proper place in the scrotal sac. A total of 18 patients had surgery before the age of 2 years, then 21 patients in the age range of 2 to 5 years, and 52 of 91patients had surgery at the age of more than 5 years. The success rate of the orchiopexy procedure performed at RSUP Dr. Hasan Sadikin in this study was 100%. (Table 4)

DISCUSSION

Results in this study showed that patients had unilateral cryptorchidism (70.3%) and bilateral cryptorchidism (29.7%). In this study, most were found to be right unilateral cryptorchidism (39.5%). This is similar to an analysis of 2150 orchiopexy data reported from seven studies in Denmark that showed the highest incidence was right unilateral cryptorchidism (46%).[10] Study from Burhan HW et al. showed the highest incidence was bilateral cryptorchidism (56,7%) and left unilateral cryptorchidism (24,3%).[5] Similar with study from Lubis SM and Pateda V, et al. showed the highest incidence was bilateral cryptorchidism (43,07%) and left unilateral cryptorchidism (29,56%).[6]

Comorbidities	Cryptorchidism			Number (n)	(%)
	Left Cryptorchid	Right Cryptorchid	Bilateral		
No Comorbid	10	13	12	35	38,5
Hydrocele	1	3	1	5	5,5
Hernia	3	2	1	6	6,6
Phimosis	3	2	-	5	5,5
Micropenis	-	1	3	4	4,4
Hypospadias	4	8	4	16	17,6
Bifid Scrotum	-	-	1	1	1,1
Retractile	3	3	-	8	8,8
Urinary Tract Infection	1	-	-	1	1,1
Testicular Hypoplasia	-	-	2	2	2,2
Ectopic Ureter	1	-	-	1	1,1
Epispadias	-	1	-	1	1,1
Testicular Torsion	-	1	-	1	1,1
Hypospadias dan Micropenis	-	2	1	3	3,3
Phimosis dan Hernia	1	-	-	1	1,1
Micropenis, Testicular Hypoplasia, and Hypospadias	1	-	-	1	1,1
Total	28	36	27	91	100

Table 3. Comorbidities in Cryptorchidism

The results of this study showed that 96.7% of cryptorchidism were diagnosed at more than 12 months of age. This indicates that there are still many cases of cryptorchidism that are late diagnosed which can result in an increased risk of complications. The study of Tasian et al. showed a 2% risk of severe germ cell loss and 1% risk of Leydig cell depletion each month if the testes remained unchanged beyond 1 year of age.[11] Decreased germ cell counts have also been reported from the age of 12 months and peaks occur at 18 months, so surgical procedures are recommended before 12 or 18 months of age.[12]

Age (years)	Number of Operations		(%)
	Success	Failure	
≤2	18	-	19,8
2 – 5	21	-	23,1
≥ 5	52	-	57,1
Total	91	-	100

Table 4. Distribution of Patients Undergoing Surgical Treatment

In this study, from the palpation examination 83 patients had isolated cryptorchidism and 8 of them were accompanied by a retractile testis, while no ectopic or atrophic testicular cases were found. The results showed that 57.1% of cases were palpable and 42.9% of cases were nonpalpable. The study from Nah SA et al. showed that 68% of cryptorchidism cases were palpable, 13% of cases were nonpalpable, 5% of cases were retractile, 7% of cases were atrophic or absent testicles, and the remaining 2% were ectopic testes.[9] Retractile testes were found in 8 cases in this study. According to Agarwal et al. 32% patients with retractile testis develops acquired cryptorchidism.[13] The testis may not be palpable due to various factors, due to its location in the abdominal cavity, inguinal canal (difficult to palpate generally in obese patients) or maybe due to atrophic or ectopic testicles.[14]

Confirmation of the diagnosis and determination of the location of the testes in RSUP Dr. Hasan Sadikin Bandung is done through an ultrasound examination or testicular exploration using laparoscopy. The results showed that most cases of cryptorchidism were found in the inguinal canal in bilateral or unilateral cryptorchidism. This is similar with study by Amooei et al. which showed that 78.6% cases of cryptorchidism

were found in the inguinal canal, 16.4% intraabdominally and there was no presence of testis (vanishing testes) for the rest of the case.[15]

The descended of testes is part of the male genital development that is regulated by a series of interactions between genetic, mechanical, and hormonal factors that also regulate the growth of the gonads, Wolffian ducts and the differentiation and growth of the genitals. Abnormalities of intersexuality (disorder of sex development) with severe virilization to failure of testicular descent can occur when there is a disturbance in the interaction of those factors.[16]

This study is a descriptive study that showed the comorbidities dan outcomes of cryptorchidism in pediatric patients at RSHS Bandung. The limitation of this study that the exact cause of cryptorchidism was not revealed because of limited cost, facilities and data. Cases of cryptorchidism accompanied by hypospadias or bifid scrotum needs to be examined thoroughly to investigate Disorders of Sexual Development. Cryptorchidism is often followed by symptoms of defects in the genital organs and several syndromes associated with chromosomal mutations and genetic variations.[16] There were 38.3% cases of isolated cryptorchidism, and the most common comorbidities were hypospadias (20.2%), followed by hydrocele, hernia, and retractile testes, each of which had 6 (6.4%) occurrence in this study. Among the 24 cases that had hypospadias and scrotum bifidum co-morbidities, only 13 cases had karyotyping and all of them were found to have 46 XY.

It is concerning that 80.2% of orchiopexy in this study was found to be performed above the age of 2 years. Cryptorchidism associated with compromised fertility is seems to be a result of multiple factors, including germ cell loss, impaired germ cell maturation, Leydig cell diminution, and testicular fibrosis. Boys with unilateral cryptorchidism have a lower fertility rate but similar paternity rate to those without cryptorchidism. Meanwhile, boys with bilateral cryptorchidism suffer both lower fertility and paternity rates. The age of surgical intervention for cryptorchidism seems to be an important predictive factor for fertility later in life. Endocrinological studies showed higher inhibin-B and lower follicle-stimulating hormone (FSH) levels in men who underwent orchiopexy at age 2 years compared with individuals who had surgery later, which is indicative of a benefit of earlier orchiopexy. Regarding preservation of fertility potential, early surgical correction of undescended testes is highly recommended before 12 months of age, and by 18 months at the latest.[17]

CONCLUSION

The orchiopexy procedure performed in cryptorchidism at RSHS was 100% success rate. Patients with cryptorchidism mostly had late diagnosed at age more than 12 months old. The most common case is cryptorchidism sinistra. It is recommended that physical examination of newborns must be done carefully, including genitalia area so that any abnormalities that were detected can be handled earlier in order to minimize the harmful risk. Late diagnosis and late orchiopexy can have long term implications for fertility and growth of the testes. Therefore, ideally the procedure needs to be carried out before one year of age ideally before 12 months of age, and by 18 months at the latest.

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