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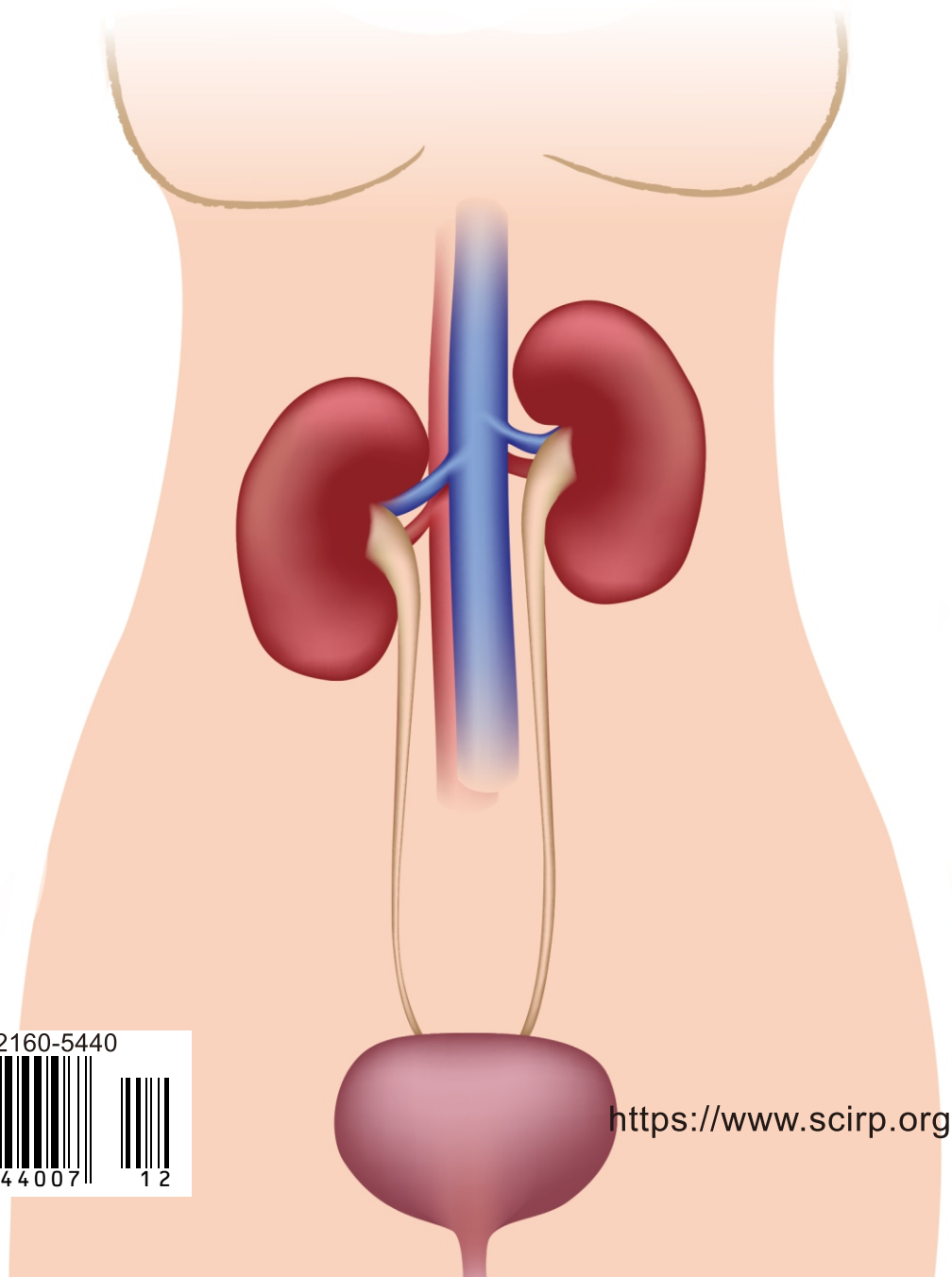

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Prevalence and Clinical Anatomic Characteristics of Cryptorchidism in Schools in the City of Conakry

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Abstract

Objective: To report the prevalence, anatomic and clinical characteristics of cryptorchidism in schools in the city of Conakry. **Material and Methods:** We carried out a 6-month cross-sectional study which took place in 32 public and private general education establishments in the city of Conakry. Each student boy was interviewed and examined; some information was collected from parents over the phone. **Results:** The prevalence of cryptorchidism was 3.6%. It was higher among children of mothers who were not in school, and among those who attended public institutions. The average age of students with cryptorchidism was 9.84 ± 2.33 years, with extremes of 4 and 17 years. 85% of these pupils were born in a care structure (CHU, municipal hospital, health center). Cryptorchidism sat on the right in 64 cases (37.4%), on the left in 44 cases (25.7%), it was bilateral in 63 cases (36.9%). The testicle was palpable at the inguinal level in 77 cases or 45% of the cases. **Conclusion:** Many school children have cryptorchidism at advanced ages, thus exposing them to the risk of complications (infertility and malignant degeneration of the testicle). Raising awareness among the various players could reverse this trend.

Keywords

Cryptorchidism, Prevalence, Pupils, School

1. Introduction

Cryptorchidism is the most common genital defect in boys [1]. It indicates the presence of a testicle, spontaneously and permanently, outside the scrotum at

any point of its normal migration path [2].

It is an obvious pathology, clinically detectable in front of the vacuity of one or both purses. Often neglected by children and their parents in our country due to a lack of information or difficulties accessing healthcare facilities, cryptorchidism can lead to sometimes dramatic consequences when it is not taken care of early. Its main complications are the risk of infertility, malignant degeneration and torsion of the testicle [1] [3].

It is to minimize this risk that most teams currently recommend lowering an undescended testicle at the age of 1 year [4] [5] [6], from which testicular migration is no longer possible spontaneously [1].

In many countries, cryptorchidism is treated at late ages, due to diagnostic delay, as evidenced by school prevalence surveys.

In Turkey in 1996, Simsek [7] noted a prevalence of 1.43% of cryptorchidism in schools.

In Nigeria in 2001 in Anambra state, Okeke [8] found a 0.82% prevalence of cryptorchidism in elementary school boys.

In our daily practice, it is not uncommon to find this anomaly in adolescents and adults. So we conducted this study in order to report the prevalence and anatomo-clinical characteristics of cryptorchidism in schools in the city of Conakry. This will ultimately enable us to sensitize populations and practitioners on the importance of early treatment.

2. Material and Methods

This was a descriptive cross-sectional study that took place in two municipalities (Matam and Dixinn) of Conakry town, capital of the Republic of Guinea. At the time of the study, the municipality of Dixinn had 134 primary and secondary schools, public and private, and that of Matam 186 schools, making a total of 320 schools. The total number of pupils enrolled in the two municipalities was 93,019, of which 47,426 were boys. We randomly chose 10% of the total school population in each municipality. For each school, we randomly selected 10% of the total student population. This allowed us to obtain a sample of 4750 students.

The study was sponsored by the national school health service, which issued us a mission order with which we went to the various schools. We carried out a large information of the teachers, the pupils and their parents on the objectives and the progress of the study.

Only boy students whose parents freely agreed to participate in the study were included in the study. It should be noted that the study took place during the Ebola virus crisis, causing reluctance in some parents of students.

The students were examined by two doctors enrolled in the Urology Specialist Studies Diploma in a room equipped with an examination table for the occasion. Palpation of the scrotum and groin was performed on confident children who were placed in the supine position. The appearance of the scrotum was noted.

When the testicle was absent from the scrotum, it was carefully searched for by inguinal and iliac fossa palpation. If the testicle was palpated in the groin, it was moved by gentle traction towards the bursa to eliminate an oscillating testicle. Thus, the diagnosis of cryptorchidism was made when the testis was not found and when the testicle palpated in the groin could not be drawn into the bursa. A general exam had completed the examination of the bursa and penis, looking for an associated abnormality.

The variables collected for each child were age, educational level of the child, place of birth, educational level of the mother, profession of the father and for pupils with cryptorchidism, the side of the anomaly, the topography of the undescended testicle, and associated anomalies were noted.

It should be noted that some children were unable to provide certain data, including their place of birth, their mother's level of education and whether they were informed of their illness. In these cases, these information were taken from the parents on the telephone.

The data were analyzed on Epi Info in its 7.1.3.3 version.

3. Results

We examined a total of 4750 students, of which 171 had cryptorchidism, a prevalence of 3.6%. This prevalence was variable according to certain variables (Table 1).

The mean age of students with cryptorchidism was 9.84 ± 2.33 years, with extremes of 4 and 17 years. The most represented age group was 6 - 10 years old (Table 2).

Regarding the place of birth of students with cryptorchidism, the CHU was the most reported with 42.6% of cases (Figure 1).

We noted that 12% of the parents were already informed of the anomaly and claimed to have consulted a specialist but finally withdrew due to the high cost of care.

According to the side, the anomaly was located on the right in 64 cases (37.4%), on the left in 44 cases (25.7%), it was bilateral in 63 cases (36.9%).

Table 1. Prevalence of cryptorchidism according to certain variables.

Variables		Number of children with cryptorchidism	Total	Prevalence (%)
Mother educational level	Un schooled	118	2520	4.68
	Pre-university	49	1968	2.88
	University	4	532	0.75
Type of school frequented	Public	149	2977	5
	Private	22	1773	1.24
Child educational level.	Primary	157	4177	3.75
	Secondary	14	573	2.44

Table 2. Distribution of students with cryptorchidism according to age groups.

Age (Revolved years)	Effectif	Percentage
1 - 5	4	2.3
6 - 10	143	83.6
11 - 15	21	12.3
≥16	3	1.8
Total	171	100

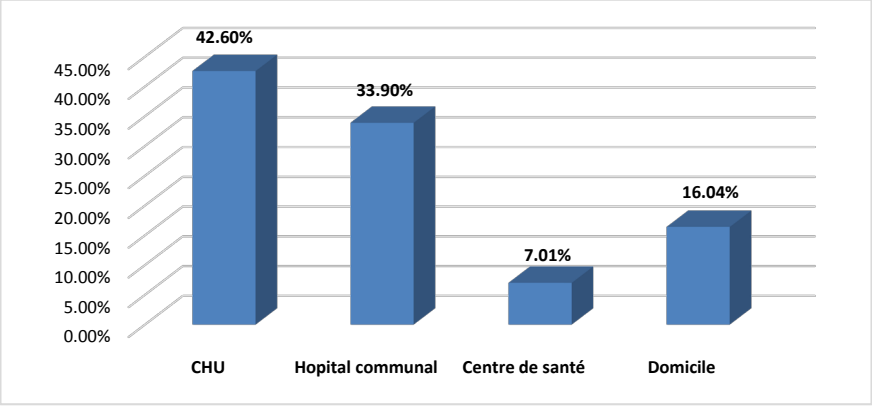

Figure 1. Distribution according to the place of birth and children with cryptorchidism.

According to the topography, the testicle was palpable at the inguinal level in 77 cases or 45%: at the root of the purse in 12 cases, *i.e.* 6.4%.

Other abnormalities were associated with cryptorchidism in some students. These were 12 cases of contralateral hydrocele, 7 cases of varicocele, 3 cases of medium penile hypospadias, 2 cases of balanic hypospadias and 4 cases of phimosis.

Of the 171 students with cryptorchidism, only 29 cases (16.95%) were able to be managed in our service.

4. Discussion

The prevalence of cryptorchidism in school-aged children varies widely in the literature. One of the highest rates was found in Denmark with a prevalence of 7% in boys aged 6 to 16 [9]. It was 2.1% in boys aged 6 to 12 years in a Jordanian study [10], while it was 1.41% in a Senegalese study reported by Gueye [11]. In our survey, we found a prevalence of 3.6%. However, we did not consider the existence of acquired cryptorchidism, which could overestimate the prevalence of true cryptorchidism in the study. Indeed, at school age, a testicle normally descended during the first year of life can ascend and secondarily fix in a high position, producing acquired cryptorchidism [1]. Hack noted a prevalence of acquired cryptorchidism of 2.2% in boys aged 6 to 13 [12].

The average age of students with testicular dystopia in our study was 9.84 ± 2.33 years with extremes of 4 and 17 years. The age of discovery advanced in our

work, as in many African series [2] [13] [14], exposes these children to a risk of infertility, and degeneration of the testicle.

The low socio-economic level has been identified as a major risk factor for diagnostic delay [15], which is probably due to the parent's lack of information and their difficulties in accessing medical structures.

Even if we did not study the standard of living in this study, it still appears that the prevalence of cryptorchidism is higher in the group of children whose mothers did not attend schools and in those who are attending public educational institutions.

It should also be noted that in 12% of the cases, the parents were already informed of the anomaly but could not cope with the high cost of care in health facilities, where they do not benefit from any social assistance.

In addition, 85% of children with cryptorchidism were born in a health facility, half of them in a CHU.

This should challenge health providers (midwives, obstetricians, pediatricians) as long as the systematic examination of the newborn in the delivery room should make it possible to detect the anomaly which is easy to diagnose [2] in the case of vacuity one or both testicle purse. Once the diagnosis has been made, children should be systematically referred to specialists for optimal management.

We notified 63.1% of unilateral cryptorchidism, with a predominance on the right side which represented 37.4% of the cases; these results corroborate the data in the literature [1] [2] [11].

The bilateral nature of the anomaly as it is the case in 36.4% of cases in our work increases the risk of infertility [16]. Lee *et al.* [16] reported a 59% male infertility rate in patients who underwent testicular lowering during childhood for bilateral cryptorchidism.

Bilaterality may be a factor in the early diagnosis of the anomaly for some authors [17].

In about 80% of cases, the cryptorchid testis is palpable [1]. The 51% rate of palpable testes in our survey is probably underestimated. In fact, the conditions of the investigation make it difficult to obtain good muscle relaxation in the child during the examination [11]. It should also be noted that on a single examination it is difficult to determine whether a palpable testicle at the root of the bursa (12 cases in our investigation) is cryptorchid or oscillating. If in doubt, the children should be seen again at other visits to determine the exact location of the testicle [15].

An associated penile anomaly would be a factor of consultation at a younger age [15]. In fact, the penis abnormality may be easier to detect by parents and may cause more concern for parents, who would request a quick consultation.

We noted 5 previous cases of balanic and penile hypospadias in our series. The fact that these hypospadias are minor could explain the possible lack of concern of the parents.

Despite the awareness, we noted that only 16.95% of children with the anomaly could be cared for in our department or in the pediatric surgery department. This is certainly linked to the extreme poverty which considerably limits access to healthcare in our country.

The main limitation of this study is that acquired cryptorchidia was not considered. We did not have access to the student health record to know the position of the testicle at birth or in the first year of life.

5. Conclusion

Our work shows that many children have cryptorchidism at advanced ages. The prevalence was higher among students born in health structures, whose mothers were not solarized and who attended public school. To reverse this trend, it will probably require an effort to raise awareness at all levels, in particular midwives, pediatricians, but also education and public information. It is also necessary to create facilities for access to care for disadvantaged families.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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Clinico-Biological Profile of the Azoosperm Patient at the Urology and Andrology Department, Conakry University Hospital

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Abstract

Objective: Azoospermia is one of the most important causes of couple infertility. The objective of our study is to report the clinical-biological profile of the azoosperm patient to the Urology-Andrology Department of the Conakry University teaching Hospital. It aims to take stock of the diagnostic management of azoospermia at this time where the world scientific community seems to be turned towards the intracytoplasmic sperm injection in the treatment of men with severe spermiological dysfunction. **Patients and Method:** This was a descriptive retrospective study lasting 12 months from January 1 to December 31, 2015. It collected 151 patients out of a set of 544 follow-ups for desire to have children. Were included the patients whose files contained all the information of the clinical observation (general information, reason for consultation, evolution, history, data of the physical examination) and a paraclinical assessment consisting of the FSH level and two spermograms spaced three months, confirming the diagnosis of azoospermia. **Results:** The mean age was 36.4 years with extremes of 23 and 56 years old. Urogenital infections (36.4%) followed by a notion of inguinal surgery had been the main patients' history. Primary infertility accounted for 76.8% of cases. The mean duration of infertility was 6.5 years with extremes of 2 and 19 years. Azoospermia affected 27.76% of patients who consulted for the desire to have a child. It was judged secretory in 59.6% of cases, excretory in 25.8% of cases, and undetermined in 14.6% of cases. Varicocele was the main associated abnormality (46.3%) followed by testicular hypotrophy (36.4%). Neisseria Gonorrhoeae was the most common germ in sperm culture (21.7%). Chlamydia serology was positive in 21.7% of patients. **Conclusion:**

Azoospermia affects a non-negligible proportion of men admitted by consulting for desire to have a child in our context. Strengthening the diagnostic and therapeutic arsenal is necessary to improve the care of affected patients.

Keywords

Male Infertility, Azoospermia, Conakry

1. Introduction

Azoospermia is the complete absence of sperm in the ejaculate [1].

It affects less than 1% of men in the general population either 5% to 15% of infertile men, and is one of the most serious causes of couple's under fertility [2] [3] [4]. Depending on the mechanism, a distinction is made between secretory azoospermia (non-obstructive) resulting from a defect in testicular sperm production, and excretory (obstructive) azoospermia linked to the existence of an obstacle on the genital tract. The combination of these two types of azoospermia is possible (mixed azoospermia) [1]. The management of azoospermia has made significant progress due to recent advances in assisted reproduction techniques and the breakthrough in the technique of intra-cytoplasmic sperm injection. It requires careful exploration of the azoosperm patient by collecting some clinical data, spermiological, hormonal, ultrasound, genetic and histological. In developing countries, obtaining these various parameters is still not possible, which makes treatment of azoospermia a constant challenge for the practitioner.

The objective of this study is to report the clinical and biological profiles of azoosperm patients diagnosed as part of their follow-up for desire for paternity in our department.

2. Patients and Method

The study was retrospective of 12 months, from January 1 to December 31, 2015. It focused on azoosperm patients who consulted during this period for desire for a child, after a minimum period of one year of married life, punctuated by satisfactory sexual intercourse without any notion of contraception. We included the patients whose files contained all the information of the clinical observation (general information, reason for consultation, evolution, history, data of the physical examination) and a paraclinical assessment consisting of the FSH level and two spermograms spaced three months.

The clinical parameters studied were age, duration of infertility, the susceptible history (trauma of bursa, inguinal surgery, uro-genital infections) alcoholism, smoking and associated physical abnormalities clinically detected and confirmed by ultrasound.

In addition to the spermogram made in the standardized procedure of WHO [5], the biological balance sheet contains a spermoculture, chlamydia serology, a hormonal balance (FSH, testosteronemia) and a seminal plasma examination

that has essentially taken into account the assay of the three main specific markers of epididym (*α*-glucosidase), prostate (citric acid) and seminal vesicles (fructose). The classification according to the type of azoospermia was essentially based on the rate of FSH and the testicular hypo/atrophy detected by physical examination or ultrasound for a testicular volume less than 16 ml [6]. So the excretory azoospermia was retained for a testicular volume superior or equal to 16 ml and a normal FSH rate; That of secretory was retained for, the testicular volume less than 16 ml and/or high or low FSH rate [7]. Our data were collected on a survey sheet after a manual analysis, then entered and analyzed by the Epi Info software in version 7.2.1.

3. Results

151 patients constituted the size of our sample. The average age of patients was 36.4 years old. (**Table 1**) The average duration of infertility was 6.52 years old (**Figure 1**). For 76.8% of patients (n = 116) it was primary infertility and 23.2% of cases (n = 35) of secondary infertility. Urinary infections in 36.4% of cases (n = 55) have dominated patients histories, followed by inguinal surgery in 11.9% of cases (n = 18) (**Table 2**). An active unweaned smoking has been noted in 40.3% of patients (n = 54) and alcohol consumption was effective in 31.3% of cases (n = 42). Varicocele in 62.91% (n = 95) was the main associated physical abnormality. It was unilateral left in 67.37% of cases (n = 64) and bilateral in 32.63% of cases (n = 31) (**Table 3**). The assistance of the FSH realized in 142 patients was high in 58.4% of cases (n = 83). For 88.89% of patients (n = 88) testosteronemia was normal (**Table 4**). Alpha glycosidase, fructose and citric acid were the biochemical markers achieved in 25.83% of our patients (n = 39) (**Table 5**). Citric acid values were normal in all patients (n = 39) and alpha glycosidase and fructose were reduced in 30.77% (12/39) and 12.82% (5/39) cases respectively (**Table 6**). Azoospermia was secretory in 59.6% of patients (n = 90), excretory at 25.8% (n = 39) and indeterminate at 14.6% of patients (n = 22) (**Table 6**). The chlamydia serology was positive in 21.7% of cases (n = 16), the Neisseria Gonorrhoeae (n = 14) and the Escherichia coli (n = 8.) were the germs isolated with spermoculture respectively in 63.7% and 36.3% of positive cultures (**Table 7**).

4. Discussion

The infertility of a couple is a major public health problem. Studies agree that 15% of the couples have difficulty procreating in 2/3 cases, an exclusive or associated male cause [8]. In 10% to 20% of cases, the abnormality in the infertile man is azoospermia [9].

The proportion of the azoospermia in our study was 27.76% compared to all patients who consulted in the period for infertility of the couple, which calls into question the endogenous beliefs which tend to victimize women in the absence of conception within households, in our African communities [10].

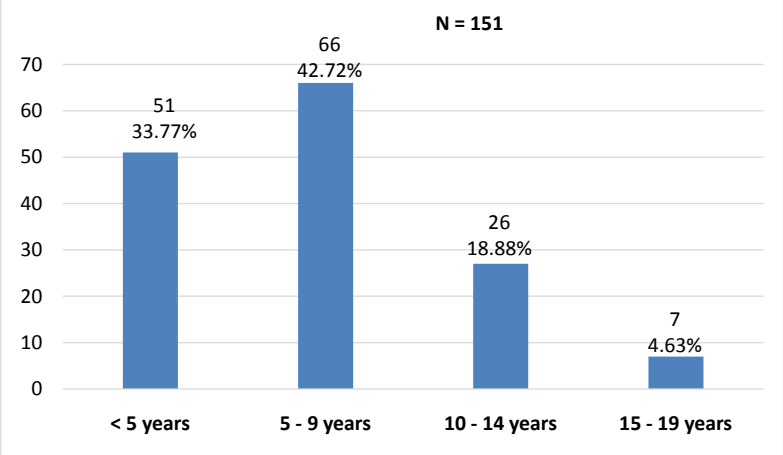

Figure 1. Duration of infertility in azoosperm patients. Average duration: 6.52 years; Extremes: 2 and 19 years.

Table 1. Age of azoosperm patients.

Age groups (years)	Effective	%
18 - 27	3	1.98
28 - 37	56	37.08
38 - 47	88	58.29
48 - 57	4	2.65
Total	**151**	**100**

Average age: 36.4 years; Extremes: 23 and 56 years.

Table 2. History of azoospermia patients.

History	Effective	%
Inguinal surgery	18	11.92
Trauma of the bursa	5	3.33
Uro-genital infections	55	36.42

Table 3. Physical abnormalities found on the patients.

Associated physical abnormalities	Effective			Total	%
	Right	*Left*	*Bilateral*		
Testicular atrophy	2	13	5	20	13.24
Cryptorchidism	7	7	------	14	9.27
Testicular hypotrophy	13	17	25	55	36.42
Epididymal cyst	1	5	1	7	4.63
Varicocele	------	64	31	95	62.91

Table 4. Hormonal assessment Result.

FSH (UI/l)	Effective	%
1.7 - 12	59	41.55
>12	83	58.45
Total	**142**	**100**
Testosterone (ηg/ml)	**Effective**	**%**
<3	3	3.03
3 - 11	88	88.89
>11	8	8.08
Total	**99**	**100**

Table 5. Result of biochemical markers assay (n = 39).

Biochemical markers		Effective	%
α-glycosidase	Normal	27	69.23
	Low	12	30.77
Fructose	Normal	34	87.18
	Low	5	12.82
Citric acid	Normal	39	100.00
	Low	0	-----

Table 6. Type of azoospermia and clinico-biological data (n = 151).

Clinico-biological data	Azoospermia type (n = 151) Total (%)			
	S* (n = 90)	E** (n = 40)	Und*** (n = 19)	
High FSH	81/83	0/83	2/83	54.97
Testicular hypo/atrophy	50/55	0/55	5/55	36.42
Cryptorchidism Epididymal cyst	13/14 0/7	0/14 4/7	1/14 3/7	9.27 4.63
Varicocele	59/95	25/95	11/95	62.91
Seminal biochemistry disturbed	0/17	17/17	0/17	11.26

S*: Azoospermia type; **E****: Excretory azoospermia; **Und*****: Undetermined azoospermia.

Table 7. Sperm culture results from azoosperm patients.

Sperm culture	Effective	%
Negative	80	78.4
Positive	22	21.6
E. coli	8	36.3
N. gonorrhea	14	63.7
Total	**102**	**100**

The average age of our patients was relatively high compared to that of azoospermia patients of developed countries that varies between 33.2 and 33.8 [11].

On average, our patients had a duration of infertility going to seven years and in the majority of cases, it was their first consultation. This time too long put to consult could explain the high average age registered by our study. For other authors, it would be the fact of the myth of the only female responsibility in the infertility of the couple [2].

The diagnosis of azoospermia is facilitated in our context by the availability and the cost more or less affordable of the spermogram. Once done, the limits of the technical equipment become a great obstacle for the practitioner to produce further explorations in order to establish etiologies with certainty. In the absence of having certainty, the search for assistance factor for the etiological diagnostic makes it possible to have an orientation.

The secretory or excretory origin of the azoospermia has rested on the criteria involving the testicular volume at the assay of the FSH rate in our patients. For KESKES, the clinical examination was sufficient to establish the secretory origin of the azoospermia based on the presence of andrologic signs (hypo/atrophy testicular, testicular epctopic) considered as revealing of a primitive disorder of spermatogenesis often associated with an increase in plasma concentrations of FSH [9].

In accordance with what is described in the literature, we found in this retrospective study, a secretory in the majority of cases. Within this patient population, 81/90 had a high FSH rate with normal testosteronemia, in almost all (88/90). Our results corroborate the fact that primitive troubles of spermatogenesis are much more common than the gonadotropic deficits in the azoospermia cases [12].

The excretory origin of azoospermia was determined in 25.8% of our patients, associated in all the cases with a history of uro-genital infections. A decrease in α-glycosidase and fructose rates was observed in 50% of these patients (n = 17) of which, 30.77% for the first (12/39) and 12.87% for the second (5/39). The genito-urinary infectuous pathology represents a large etiological group in male infertility [13]. If in the developed countries, the ABCD remains the main cause of obstructive azoospermias, in our context, the high rate of cases of non-poorly treated infections is one of the factors for acquired obstruction from the seminal path [14].

The history and physical abnormalities found in our patients are all likely to have an impact on fertility. For some just as tobacco, the link with azoospermia does not seem to be classic. Cigarette smoking has negative effects on male fertility. Recent studies showed an active transfer of several components of cigarettes through the blood-testis barrier. The presence of these components in the seminal plasma may induce a degradation of sperm parameters and nuclear quality of spermatozoa, and compromise the chances of pregnancy. [15].

On the other hand, with some such as cryptorchidia, urogenital infections and

the notion of inguinal surgery, the literature data appear to agree on the existence of a link with the occurrence of azoospermia.

Cryptorchidia is recognized as one of the main risk factors of azoospermia in 15% to 20% of cases [16].

In addition to being at the origin of azoospermia by obstruction of spermatic channels, the infections of the uro-genital paths are not only known to be at the origin of a leucospermia generating an oxidizing stress for sperm but also to be producing anti-spermaticozoid antibodies [17].

Inguinal surgery came to the second rank of the history found in our patients. Because of the presence of the spermatic cord, it is associated with any surgical act realized in this anatomical region of the body, a risk of iatrogenic damage on its elements. Two mechanisms reflect the possibility of occurrence of azoospermia in this type of patient: the damage of vascular structures resulting in ischemic orchitis, a testicular atrophy or endocrine dysfunction on the one hand and compression or even the stenosis of the canal defects on the other hand. When existing, these damage mechanisms can only compromise fertility in particular in case of bilateral surgery or pre-existing deficiency of the controalateral testis [1]. Varicocele has been the main physical anomaly found in our patients. It was dominant on the left (63.75%) and bilateral in 36.25%. The difficulty in the analysis of the results was the impossibility of determining whether it had a role in the occurrence of the azoospermia or if it were a simple association. Admittedly, the oligo-astheno-theratorspermia remains the most characteristic spermiological anomaly of varicocele, [18] but azoospermia may result from the ultimate evolution of varicocele by degradation of the spermogram [2].

The retrospective nature of this study was the main limitation. from the fact, some balance sheet, such as seminal plasma analysis and bursa ultrasound, could not be performed uniformly in all patients. Likewise, the lack of genetic tests and testicular biopsy were the source of missing and yet crucial variables in the etiological research of azoospermia.

5. Conclusion

Azoospermia is a spermiological abnormality that affects 27.76% of the men consulting in our context for child desire. It is dominated by secretory origin and the concerned patients have a profile that may be of interest to all etiologies. In the perspective of the introduction of methods of reproductive assistance, it seems imperative to strengthen the arsenal diagnosis through the development and extension of biological exploration and more efficient imaging means.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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Isolated Caeco-Vesical Fistula: A Case Report

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Abstract

Congenital uro or genito-digestive fistulas are frequently found in the context of caudal pole malformations. Isolated congenital forms without associated anorectal malformation have not been reported until now. We report the first case we know a 9 year old female child received in a pediatric surgery consultation for fecaluria evolving since birth with a permeable anus. She presented a good general condition, a good staturo-ponderal and psychomotor development, a vulva soiled by stool and a permeable anus with a tonic sphincter. A retrograde urethrocystography revealed a caeco-vesical fistula. Surgery consisted of ligation-section of the caeco-vesical septum. A follow-up urethrocystography at three months post-surgery no longer visualized the fistula. Our post-operative follow-up is 4 years. Isolated congenital caeco-vesical fistula is an unknown pathology whose late diagnosis can have serious repercussions.

Keywords

Fistula, Congenital, Isolated, Caecum, Bladder, Child, Surgery

1. Introduction

Congenital uro or genito-digestive fistulas are frequently found in the context of malformations of the caudal pole (anorectal malformation, pouch colon, dupli-

cation of the urethra, aphaly) [1] [2]. Congenital isolated forms without associated anorectal malformation such as isolated caeco-vesical fistula have so far not been reported. In acquired forms, they are of late diagnosis and are revealed by fecaluria. They are described in elderly patients and in particular pathologies such as Crohn's disease [3], colonic diverticulosis [4], caecal cancer [5], and more rarely intestinal amoebiasis [6], bladder bilharziasis [7] and intestinal hydatidosis [8], and even in these cases that caeco-vesical fistula remains exceptional. We report an observation of the first case, to our knowledge, of isolated congenital caeco-vesical fistula.

2. Observation

A 9-year-old girl is seen in a paediatric surgery consultation for fecaluria associated with a permeable anus which the parents had observed since birth. The late consultation is only justified by the pungent smell of urine soiled with almost permanent stool of the child who had her schooling interrupted. A notion of repeated urinary tract infections was signaled in her history. The girl was in good general condition, with normal staturo-ponderal and psychomotor development (Height: 145 cm/Weight: 35 kg). There was a pungent smell of urine mixed with stool and coloured conjunctiva. The examination of the perineum revealed a stool-stained vulva, a wide urinary tract and an open vaginal orifice; a permeable anus with a tonic sphincter and a palpable stool-filled rectum.

The diagnosis was obtained by retrograde urethrocystography (UCR) which revealed a caeco-vesical fistula (**Figure 1**).

Following a water-soluble enema, it was possible to rule out recto-vaginal and recto-vesical fistula. Creatininaemia was normal. A week-long preoperative hospitalization was necessary for dietary management, colic draining by enemas and bladder asepsis (antibiotic therapy). A strict 72-hour diet resulted in clear urine with no microbial germs as confirmed by cytobacteriological examination. A standard preoperative assessment was carried out (complete blood count, hemostasis assessment and blood grouping).

The treatment was surgical with a right para-rectal approach, allowing an easy access to the caecum. During the exploration, we visualized the caeco-vesical septum (**Figure 2**) with an appendix passing underneath (**Figure 3**). The procedure consisted of the ligation-section of the caeco-vesical septum and an appendectomy of necessity (**Figure 4**).

The postoperative follow-up was simple with the resumption of the intestinal transit at day 4, authorized feeding at day 5, and the removal of the urinary catheter at day 8 post-surgery. The daily diuresis was 65 ml/hour.

Anatomopathological analysis of the appendix revealed no abnormalities.

She was discharged 10 days after surgery. Clinical and biological follow-up (cytobacteriological examination of urine, creatininaemia) did not reveal any recurrence. A retrograde urethrography review at three months postoperative found no fistula (**Figure 5**). Our postoperative follow-up is 4 years.

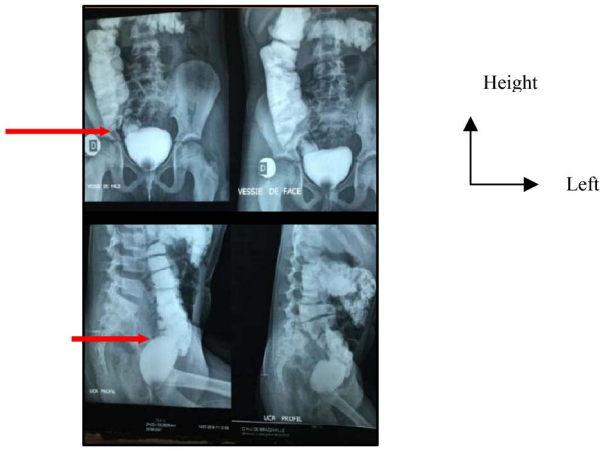

Figure 1. Retrograde urethrocystographyhighlightingthe fistula.

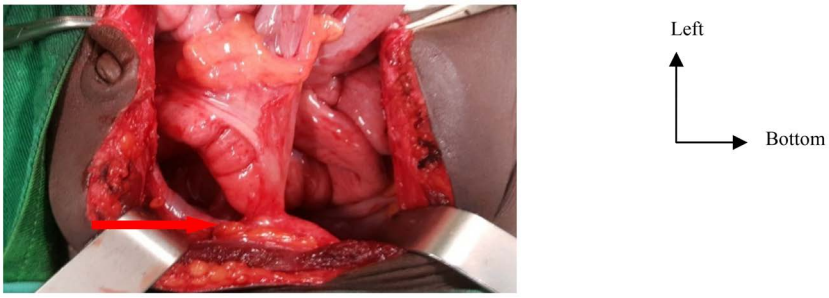

Figure 2. Caeco-vesical septum.

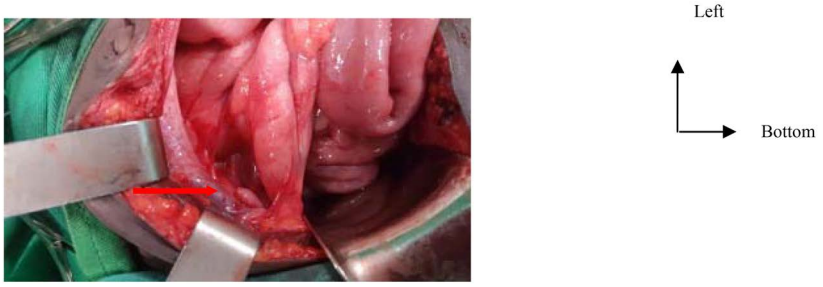

Figure 3. Appendixpassing underneath the caeco-vesical septum.

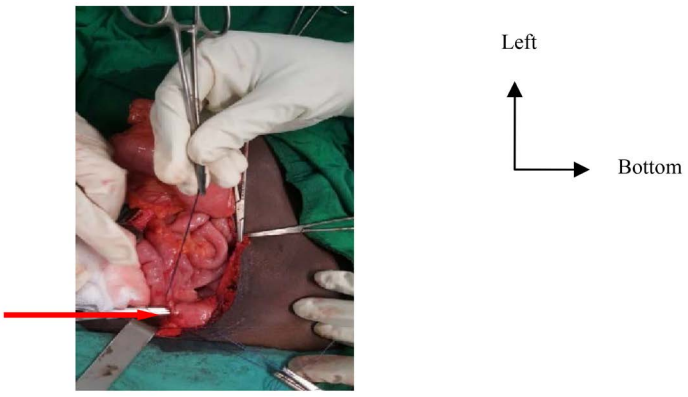

Figure 4. Sectionning of the septum and ligation of caecal and bladder banks.

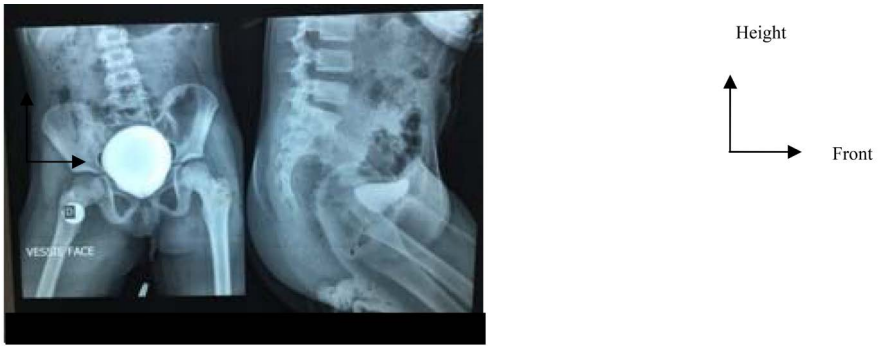

Figure 5. Retrograde urethrocystography 3 months post-surgery.

3. Discussion

Congenital uro-digestive fistulas are found in caudal pole malformations such as anorectal malformations and the pouch colon [1] [2]. Anorectal malformations are the most common congenital surgical abnormalities of the intestine. They occur in embryogenesis and in nearly 70% of cases are associated with other malformations (urogenital, musculoskeletal, cardiac, digestive and central nervous system) [9]. All forms exist, from fistula in situ to the total absence of the anus with or without urinary or genital fistula. The Peña classification [9] defines anorectal malformations as high or low depending on the position of the rectal cul-de-sac in relation to the supporting muscles and the level of a possible fistula. However, like the Krickenbeck classification [10], it does not describe a caeco-vesical fistula. This classification categorizes lesions in large clinical groups according to the location of the fistula (perineal, recto-urethral, recto-vesical, vestibular), cloacal and fistula-free lesions, and anal stenosis, as well as rare and regional variants such as the pouch colon, atresia or rectal stenosis, recto-vaginal fistula, the H fistula and others [10].

The pouch colon, in which a pocket-shaped dilation of a colon shortened by variable degrees is associated with an anorectal malformation, is a common anomaly in India. The pouch usually ends with fistula connecting to the genitourinary tract. In girls, the fistula leads either into the urethra or into the vestibule, and often a double vagina is found [1] [2]. It is currently classified as a rare regional variant of ano-rectal malformations [10]. Diagnostic confirmation is done through standard abdominal X-ray examinations without the need for enema [1] [2]. We found a permeable anus with a tonic sphincter; following a water-soluble enema it was possible to rule out a recto-vaginal or rectovesical fistula and colic pocket dilation, and hence an anorectal malformation or pouch colon. In our case, the isolated nature of caeco-vesical fistula is justified by the absence of other associated malformations and the confirmation by retrograde urethrocystography. We did not find an isolated cause of congenital caeco-vesical fistula.

Acquired entero-vesical fistulas are essentially the result of an inflammatory and infectious process of digestive origin, which will eventually develop into a

fistula in a healthy bladder. Most often the intestine responsible is either the terminal ileum or the sigmoid. The two main etiologies of entero-vesical essentially colo-vesical fistulas, are sigmoidal diverticulosis [4] and Crohn's disease [3], which are found in the elderly. Sigmoidal diverticulosis is not in itself a disease, it is a hernia of the mucous membrane through the colon muscle wall. Its symptomatic form of complicated diverticulitis accounts for 40% - 78% of the etiologies of colo-vesical fistula, with complications that can range from a simple peri-sigmoid abscess to the perforation of a diverticula in the large peritoneal cavity that causes generalized acute peritonitis, or in a neighbouring hollow organ, often giving a sigmoid-vesical fistula, or more rarely sigmoid-vaginal, sigmoid-ileal or even sigmoid-cutaneous fistula [4]. Clinical signs with type of pneumaturia and fecaluria are pathognomonic of the diagnosis. Crohn's disease is a chronic inflammatory bowel disease that can cause contiguous urinary tract damage, and responsible, though rarely, for entero-vesical fistulas [3]. In this case, we found a healthy colon. We did not perform any prior diagnostics, either urological or digestive, in the face of the findings of the radiological examinations and the difficulty of implementation in our center.

Caeco-vesical fistula in caecum cancer and intestinal amoebiasis are described in the elderly. Losco [5] describes a case of caecum cancer with caeco-vesical fistula in a 77-year-old patient with a history of endometrial cancer with hysterectomy and radiotherapy 18 years earlier. Vincent [6] describes the first case of caeco-vesical fistula due to a little-known and untreated intestinal amoebiasis in an 80-year-old patient in the form of acute peritonitis. Entero-vesical fistulas are exceptional in parasitic infections but can occur. Yddoussalah [7] describes the first case of vesico-sigmoidal fistula complicating bilharziasis in a 71-year-old patient, and Lahyani [8] the first case of vesico-sigmoidal fistula complicating intestinal hydatidosis in a 48-year-old patient. We did not perform biological tests for intestinal amoebiasis, urinary bilharziasis or hydatidosis, there being no anamnestic indication for these infections and because the symptomatology in our patient existed since birth. In urinary bilharziasis or hydatidosis, fistulas are essentially vesico-sigmoidal. Surgical treatment is done in a single operation after appropriate preparation of the colon [3] [11] except in emergency situations where a derivation in time is possible [5].

4. Conclusion

Isolated congenital caeco-vesical fistula is a little-known pathology yet of simple diagnosis and treatment. Late diagnosis has serious implications such as urogenital infections with the risk of long-term kidney failure and social isolation for the child.

Parents Inform Consent

We attest that the child's parents were informed and gave their accord for the publication of this case report.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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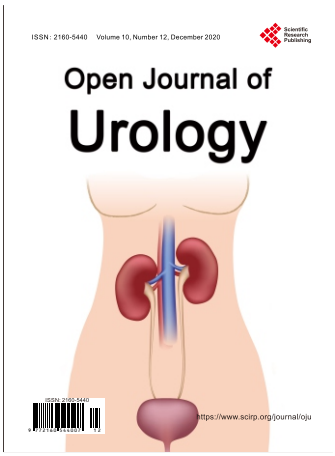

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