UNDESCENDED TESTES: PERSPECTIVE FROM A DEVELOPING COUNTRY

¹Ekenze SO, ²Esom EA, ¹Nwangwu EI

¹Sub-Department of Paediatric Surgery, College of Medicine University of Nigeria, Enugu Campus, Enugu, Nigeria ² Department of Anatomy, College of Medicine, University of Nigeria Enugu Campus

ABSTRACT

OBJECTIVE: This report details the findings at operation and challenges of managing undescended testes (UDT) in southeastern Nigeria.

METHODS: Prospective evaluation of children managed for undescended testis from January 2013 to November 2015 in Enugu, southeastern Nigeria.

RESULTS: There were 54 patients with total of 69 undescended testes (39 unilateral, 15 bilateral) involving the left side in 39 and the right side in 30 cases. The median age at operation was 5 years (Range 1 – 15 years). Three (4.35%) testes were localized at suprascrotal position, 12 (17.4%) were emergent, 33 (47.8%) canalicular, 18 (26.1%) abdominal, and 3 (4.35%) were vanishing. The volume of the localized testes ranged from 0.7mls to 8.5mls (mean 2.0mls). Epididymal anomaly was noticed in 31 (47%) with the commonest anomaly being head non-fusion. Age at operation was an independent predictor of testicular volume (OR 4.91). Single stage scrotal repositioning was achieved in 28/37 (75.7%) of cases and was not dependent on age (p=0.58). Testes localized distal to the internal ring are more likely to be mobilized to the scrotum at initial orchidopexy than those located in intraabdominal position (44/48 vs 4/18; p<0.05).

CONCLUSION: Most of the patients with UDT in our setting had surgery at a later age than recommended. The age at operation correlated with the size of the testis but did not affect the location of the testis, and the success with one-stage scrotal repositioning. Long-term follow up and evaluation is imperative.

KEY WORDS: Undescended testes; anomalies; morphology; Challenges; Developing Country

NigerJmed2018: 294-299 © 2018. Nigerian Journal of Medicine

INTRODUCTION

Undescended testis (UDT) is the commonest anomaly of male sexual development¹. It affects about 4% of full-term male newborns and 30% of preterm babies.^{1,2} The affected testes may descend into the scrotum in about 75% of full-term neonates and in about 90% of premature newborn boys in infancy, the incidence decreases to 0.8-1.2% at 1 year of age.¹⁻³ Though endocrinopathy and structural abnormalities of the gubernaculum and abdominal wall have been recognized as factors affecting descent of the testis, to a great extent, the aetiology of UDT is unclear.^{4,5}

Patients with undescended testes should be treated because of increased risk of infertility, testicular cancer, torsion and/or accompanying

Correspondence to: Dr. Sebastian O Ekenze Sub-department of Paediatric Surgery College of Medicine, University of Nigeria Teaching hospital Enugu, 400001, Nigeria Email: sebekenae@gmail.com Tel: 234-803-777-3831 inguinal hernia (>90%), as well as cosmetic concerns.^{1 - 3, 5 - 7} Management of UDT involves localization, use of hormonal manipulation, and surgical mobilization and fixation. Currently, laparoscopic treatment is the gold standard for localization and definitive treatment especially for the impalpable testes.⁸ Optimal outcome in UDT is obtained if definitive treatment is concluded before or at one-year of age.^{1-3,5-7,9}

This is premised on the preservation of spermatogonia, which is the stem cells for subsequent spermatogenesis, and reduction in the incidence of testicular cancer. Despite surgical treatment by orchidopexy, the long-term outcome may still be challenging and controversial. Findings from some previous publications indicate that in patients with UDT, the location of the testes as well as the size and the inherent morphological abnormalities may significantly impact on the treatment and the ultimate outcome.^{2,3,10}

In some developing countries, delayed presentation and inadequate facilities for proper management are challenges that may negatively affect the outcome.¹¹⁻¹³ In addition, there is scant data on the gross morphology of the testes as well as the implication of this on the treatment and eventual outcome in this setting.

This report looks at the morphological characteristics of the testes in children operated for undescended testes in two referral centres in southeastern Nigeria. The focus is on the age at orchidopexy, findings at operation, and challenges of management.

PATIENTS AND METHODS

At the University of Nigeria Teaching Hospital and the Mother of Christ Specialist Hospital in Enugu southeast Nigeria, the paediatric surgery unit undertakes the management of UDT. Patients with UDT are referred to these centres from the surrounding primary and secondary health facilities in the southeastern region of Nigeria, and from the paediatric department of the two hospitals. The estimated population of the region served is 16 million persons, and the birth rate is 35.5-births/1000 population.¹⁴ On presentation, the patients are evaluated clinically by history and physical examination with emphasis on determining the location of the testes and associated anomalies. In our setting, facilities for laparoscopy and Magnetic Resonance Angiography are not available. After the clinical assessment and notwithstanding the success with preoperative localization of the affected testes, the patients are optimized for open orchidopexy. Under general anaesthesia final attempt to preoperatively localize the testes before orchidopexy is done.

Patients were recruited for this study from January 2013 following appropriate consent. During evaluation, data collected were age at presentation, side affected, preoperative location of the affected testes, and the presence or otherwise of patent processus vaginalis. At operative treatment, data on the age at operation, definitive location of the testes, size of the testis, consistency of the testis, presence and type of epididymal anomaly, patency of the processus vaginalis, adequacy of cord length, and the lowest point of fixation at the initial surgery were collected. Also documented were the distal gubernaculum attachment, procedure undertaken and the number of procedures to get the affected testes to the scrotal fundus and the duration of follow up. The dimension of the testis (width, height, length) was measured in millimeters with a measuring tape, and the volume calculated based on the formula.¹⁵ width x height x length x 0.71. The consistency of the testis was described as firm, or soft, and if blind ending vas deferens and vessels was found, the term vanished testis was applied. Epididymis was described as normal if it is well aligned to the body of the testis, type 1 anomaly if there is head nonfusion, type 2 anomaly if there is tail non-fusion, and type 3 anomaly if there is complete nonfusion. Spermatic cord length was considered adequate if the mobilized testis could achieve scrotal repositioning, and short if the testis could not reach the scrotum. During the procedures, suspicious-looking testes were biopsied for histopathology to exclude neoplastic transformation. The lead author undertook all the operations

DATA ANALYSIS

Statistical package for Social Sciences (SPSS 15.0 version, SPSS Inc, Chicago, Ill) was used for data entry and analysis. Results were expressed as percentages, or mean. Data were analyzed by chi-square test and regression analysis as appropriate. In all, the significance level was set to P < 0.05.

RESULTS

There were 54 patients with a total of 69 undescended testes (unilateral in 39 cases; bilateral in 15 cases). The left testis was affected in 39 cases, and the right testis in 30 cases. Although the anomaly was noticed at a median age of 6 months (range 1 month – 10 years) the patients presented to our service at a median age of 1.5 years (range 6 months – 15 years).

Findings at operation

Summary of the findings at operation is shown in Table one. The median age at operation was 5 years (Range 1 – 15 years). Only 3 (4.3%) patients had orchidopexy at one year of age. At operation the testes were located at the suprascrotal position in 3 (4.4%), emergent in 12 (17.4%), canalicular 33 (47.8%), and abdominal in 18 (26.1%). Blind-

ending vas and testicular vessels (vanished testes) was found in 3 (4.4%) cases. The consistency of the 66 localized testes was firm in 44 (66.7%) cases and soft in 22 (33.3%). Of the testes localized in the extra-abdominal position, 70.8 % (34/48) had firm consistency compared to 55.6% (10/18) of the testes in intraabdominal location (p=0.379). The

volume of the 66 testes ranged from 0.70mls to 8.5mls (mean 2.0mls). On regression analysis age at operation (p=0.005) was the only significant predictor of testicular volume with odds ratio of 4.91. The other factors analyzed were location of the testes at operation (p=0.183), and presence of epididymal malformation (p=0.270).

Table 1: Showing summar	v of the findings on	the 66 testes at operation
0		

Age at Operation <i>n</i> Operative location*Mean Volume					Consistency	Site of Fixation at initial surgery				
SS EG CC	ABDFirm	Soft	Scrotal		Inguinal					
1 – 5 years	37	2	7	20	8	1.5ml (0.7-3.2)	26	11	28	9
6 - 10 years	22	1	3	9	9	2.1ml (1.0-7.1)	12	10	16	6
> 10 years	7	0	2	4	1	4.8ml (2.4-8.5)	6	1	4	3
Total	66	3	12	33	18		44	22	48	18

* SS (Suprascrotal); EG (Emergent); CC (Canalicular); ABD (Abdominal)

Table two compares the volume of the undescended testes in the various age groups with the volume of normally descended testes.¹⁶ Overall, 31 (47%) of the localized testes had epididymal anomaly, while there was associated patent processus in 43 (65.1%) of the testes.

Table 2: Comparison of the mean testicular volume in the 66 undescended testes with volume of normally descended testes measured with Prader Orchidometer¹⁶

Age range	Mean Volum	e of Testes	
Our cases of Undescended testes	Normally	Descended Testes	
1-5 years	1.5ml	1.7ml	
6-10 years	2.1ml	2.4ml	
>10 years	4 .8ml	8.7ml	

Table three shows the age at operation and the distribution of the types of epididymal anomaly and patent processus among the 66 testes. Epididymal anomaly was not predicted by the location of the testes (p=0.743), or the size/volume of the testes (p=0.296). Of the cases with associated patent processus, 41 (95%) involved testes localized distal to the internal ring (p<0.001). None of the 66 localized testes showed features of neoplasm.

Table 3: Showing the distribution of the types of epididymal anomaly, and patent processus among the 66 testes

Age at operation <i>n</i> Epididymal anomaly [#] Patent processus (%)					
	None	HnF	TnF	CnF	
1 –5 years	37	18	7	4	824 (64.9%)
6 - 10 years	22	13	4	2	3 13 (59.1%)
>10 years	7	4	2	1	0 6 (85.7%)
Total	66	35	13	7	1143 (65.1%)

HnF (Head non-fusion); TnF (Tail non-fusion); CnF (Complete non-fusion)

Operative treatment

The spermatic cord was of adequate length in 48 (72.7%) of the localized testes, and short length in 18 (27.3%). The former cases had single stage orchidopexy to the scrotum, while the latter required 2-stage procedure to achieve orchidopexy to the scrotum. Single stage scrotal repositioning was achieved in 28/37 (75.7%) of cases operated at age one year to five years compared to 16/22 (72.7%) at age 6 years to 10 years, and 4/7 (57.1%) at more than 10 years of age (p=0.58). The testes localized distal to the internal ring are more likely to be mobilized to the scrotum at initial orchidopexy than those located in intraabdominal position (44/48 vs 4/18; p<0.001)

Follow up

The average duration of follow up of these cases was 18 months (range 6 – 38 months). There were two cases of atrophy noticed twelve to eighteen months after initial orchidopexy. This occurred among the patients with short cord length.

DISCUSSION

This study has shown that a significant majority of our cases had late orchidopexy. The current standard of care in undescended testes is for orchidopexy to be undertaken between the ages of 6 months and 12months.^{1-3,5-7,9} This has been shown to maximize the future fertility potential and decrease the risk of testicular cancer. Despite this well-established guideline, studies have shown that most ochidopexies are not performed at the proper time in many developing countries.¹¹⁻¹³ The reports from some developed countries^{2, 9, 17, 18} also indicate that some cases of UDT had surgery at a later age than recommended. In our setting, this delayed treatment maybe caused mainly by delayed referral of these patients that lead to delayed presentation to the mainstream hospitals and older age at definitive treatment. Similar experience has been reported for some other congenital anomalies.^{11, 12, 19} Additional factor may be lack of primary care continuity, and parental ignorance.¹¹ Feasible initiative to ensure earlier primary care referral directly from the routine postnatal check to appropriately staffed and equipped centres may improve time to diagnosis and treatment.

A number of the findings at operation in the present report may be similar to the findings documented in some previous studies. Firstly, on the location of the undescended testes, this study and some published literature indicate that the

inguinal canal is the commonest site.^{2, 7, 9, 20} The reason for the arrest of most UDT in this location is unknown. However, studies^{3, 21} suggest that the complexity of the second phase of testicular descent may be contributory. In this phase also known as the inguinoscrotal phase, and which takes place between 25 and 35 weeks' gestation, androgens acting indirectly via the genitofemoral nerve direct the migration of the gubernaculum and testis from the inguinal region to the scrotum. Due to the complexity of this phase of testicular descent, it may make the process more susceptible to endocrine disrupters and the descent of the testis more vulnerable to arrest. Secondly, the volume of the testes in this report despite the limitation inherent in our measurement technique can be compared with established data in this domain.^{15,22,23}

Though increasing testicular size would be expected with growth and advancing age as our study and some previous studies^{22, 23} show, there are indications that the size of the undescended testes may be smaller than the expected size in descended testes.^{16, 22, 23} As table 2 has shown, this may more pronounced at puberty. Thirdly, the proportion of vanished testes in this study is similar to 5% reported in literature.^{24, 25} The significance of these tissue nubbins that constitute the vanishing testis syndrome in future tumor development is currently unknown, but their excision is recommended.²⁵ Finally, despite the age at operation, none of our cases had features of neoplasm unlike some previous reports^{5, 6, 26} that documented clinical neoplasm. This may be related to the small number of our cases. It has been noted that the incidence of malignancy increases with advancing age especially postpubertal.²⁶ Our findings notwithstanding, it is instructive to ensure long-term follow up of the patients to exclude the development of neoplasm.

Different degrees of fusion anomalies of the epididymis are reported in 36 to 79% of boys with congenital undescended testis^{10, 20, 27, 18}, and the finding of such associated anomalies in the present report corresponds to this. The significance of these anomalies is unknown and the reported incidence may be related to index of suspicion of their presence during the operative treatment of UDT.²⁷ Previous studies^{27, 28} have however reported similar anomalies in conditions with patent processus such as hydrocele, but surmised that this elicited no significant

abnormalities in germ cells per tubule. Despite our findings, it may be instructive to have long-term outcome data on the size of testis and epididymal separation to obtain valid conclusions.

The treatment of UDT may involve the use of hormonal manipulation, or surgical mobilization and fixation.^{1, 3, 7, 9} Currently, AUA guideline recommends laparoscopic procedure for localization and definitive treatment especially for non-palpable testes.^{1,7} In our study, all the cases were managed with open surgical mobilization and fixation because facility for laparoscopic surgery was not available in the period of this study. Despite this, more than two-thirds of the UDT achieved scrotal position with the initial orchidopexy. While this may be comparable with the results from better-equipped settings^{2,3,5,7,9,22,29}, it is pertinent to note that intra-abdominal location of the testis may significantly reduce the chance of scrotal repositioning at initial orchidopexy, In patients with intra-abdominal testes, a study³⁰ has shown that the presence of patent processus and the use of laparoscopic techniques might improve success with mobilization of the testis to the scrotal position. There is controversy on the best treatment option for cases that present after puberty. On one hand, most authors^{5, 6, 9, 26} have recommended orchiectomy for post-pubertal cases, citing the reported increased risk of malignancy. On the other hand, some authors^{31, 32} advocate that treatment of such cases be individualized with orchidopexy undertaken in cases without intraoperative evidence of malignancy. The premise for the latter option is unclear, but they consider orchidopexy with regular scrotal examination a suitable treatment options for post-pubertal cases of UDT without malignant changes.

Our duration of follow-up is largely insufficient to come to any conclusion except on the immediate post-surgical atrophy. Long-term follow up of cases of UDT who underwent orchidopexy is useful for appropriate counseling and monitoring for early detection and treatment of cases that may develop cancer, testicular atrophy or infertility. This has become imperative in view of the finding that successful scrotal repositioning of the testis may reduce but does not prevent potential longterm issues of infertility and testis cancer.^{1,22}

LIMITATION OF THE STUDY

The small number of cases was a major limitation of this study. This did not allow for adequate comparative with published large series. Also the duration of follow-up is largely insufficient to come to any significant conclusions on the effect outcome of operative treatment. The findings on the size of testis and epididymal separation may not engender valid conclusions without longterm outcome data.

CONCLUSION

In our setting, most of the patients with UDT had surgery at a later age than recommended. These testes at operation were localized frequently in the inguinal canal. The age at operation correlated with the size of the testis but did not affect the location of the testis, or success with one-stage scrotal repositioning. Intra-abdominal location of the testis may significantly reduce the chance of scrotal repositioning at initial orchidopexy. Developing an efficient referral system and population-based public enlightenment programme may engender timely orchidopexy, while long-term follow up and evaluation may be imperative to monitor the abnormalities and for potential infertility and testicular cancer.

REFERENCES

- Kolon TF, Herndon CD, Baker LA, Baskin LS, Baxter CG, Cheng EY, et al. Evaluation and treatment of cryptorchidism: AUA guideline. J Urol 2014; 192(2): 337 – 45.
- 2. Nah SA, Yeo CS, How GY, Allen JC Jr, Lakshmi NK, Yap TL, et al. Undescended testis: 513 patients' characteristics, age at orchidopexy and patterns of referral. Arch Dis Child 2014;99(5):401-6.
- Hutson JM, Balic A, Nation T, Southwell B. Cryptorchidism. Semin Pediatr Surg 2010; 19(3):215-24.
- 4. Lacerda HM, Richiardi L, Pettersson A, Corbin M, Merletti F, Akre O. Cancer risk in mothers of men operated for undescended testis. PLoS One 2010; 5(12):e14285. doi:10.1371/journal. pone.0014285.
- Chan E, Wayne C, Nasr A. Ideal timing of orchiopexy: a systematic review. Pediatr Surg Int 2014; 30(1):87 – 97
- Pettersson A, Richiardi L, Nordenskjold A, Kaijser M, Akre O. Age at surgery for undescended testis and risk of testicular cancer. N Engl J Med 2007; 356(18): 1835 – 41.
- Sapin E. Cryptorchidism: Guidelines for surgical management. Arch Pediatr 2014; 21(1):113-7.

- Mehendale VG, Shenoy SN, Shah RS, Chaudhari NC, Mehendale AV. Laparoscopic management of impalpable undescended testes: 20 years' experience. J Minim Access Surg 2013;9(4):149–53
- Hensel KO, Caspers T, Jenke AC, Schuler E, Wirth S. Operative management of cryptorchidism: guidelines and reality--a 10-year observational analysis of 3587 cases. BMC Pediatr.2015; 15: 116. doi: 10.1186/s12887-015-0429-1.
- 10. Rachmani E, Zachariou Z, Snyder H, Hadziselimovic F. Complete testis-epididymis nonfusion anomaly: a typical association with cryptorchid testis. Urol Int 2012; 89(3): 355–7.
- Ekenze SO, Ikechukwu RN, Oparaocha DC. Surgically correctable congenital anomalies: prospective analysis of management problems and outcome in a developing country. J Trop Pediatr. 2006; 52 (2): 126–31
- 12. Ahn H, Lee HE, Park K, Choi H. Reasons for delayed orchiopexies in a korean tertiary care hospital. Korean J Urol. 2014;55(1):69–73.
- Agossou-Voyeme AK, Fiogbe MA, Goundete J, Hounnou GM, Hodonou R. Paediatric urologic pathologies at National Teaching Hospital in Cotonou: Aetiological and therapeutic aspects. Afr J Paediatr Surg 2013; 10(2): 135 – 9.
- 14. 2006 Population Census of the Federal republic of Nigeria. Analytical report at the National level. National Population Commission, 2011.
- 15. Sakamoto H, Saito K, Oohta M, Inoue K, Ogawa Y, Yoshida H. Testicular volume measurement: comparison of ultrasonography, orchidometry, and water displacement. Urology 2007;69(1):152–7.
- 16. Goede J, Hack WW, Sijstermans K, van der Voort-Doedens LM, Van der Ploeg T, Meij-de Vrie A, et al. Normative values for testicular volume measured by ultrasonography in a normal population from infancy to adolescence. Horm Res Paediatr 2011; 76(1):56-64. doi:10.1159/ 000326057.
- 17. Hrivatakis G1, Astfalk W, Schmidt A, Hartwig A, Kugler T, Heim T, et al. The timing of surgery for undescended testis a retrospective multicenter analysis. Dtsch Arztebl Int 2014; 111(39): 649 57.
- 18. Bradshaw CJ, Corbet-Burcher G, Hitchcock R. Age at orchidopexy in the UK: has new evidence changed practice? J Pediatr Urol 2014; 10(4):758–62.
- Ekenze SO, Ngaikedi C, Obasi AA. Problems and Outcome of Hirschsprung's Disease Presenting after 1 year of age in a Developing Country. World J Surg 2011; 35 (1): 22 – 26

- 20. Meij-de Vries A, Hack WW, Heij HA, Meijer RW. Perioperative surgical findings in congenital and acquired undescended testis. J Pediatr Surg 2010; 45(9):1874 – 81
- 21. Hutson JM, Thorup J. Evaluation and management of the infant with cryptorchidism. Curr Opin Pediatr 2015; 27(4): 520 4.
- 22. Rossi V, Sartori A, Bordin G, Parolini F, Morandi A, Arnoldi R, et al. Cryptorchidism: mediumand long-term follow-up. Minerva Pediatr 2013 Jun; 65(3): 261 – 9.
- 23. Jedrzejewski G, Wozniak MM, Madej T, Kryza R, Zielonka-Lamparska E, Wieczorek AP. The differences in testicular volumes in boys 8-36 months old with undescended, retractile and hydrocele testis--usefulness of scrotal screening ultrasound. Early Hum Dev 2012; 88(3): 185-9.
- 24. van Brakel J, Dohle GR, de Muinck Keizer-Schrama SM, Hazebroek FW. Different surgical findings in congenital and acquired undescended testes. BJU Int 2012; 110(8 Pt B):E387-91. doi:10.1111/j.1464-410X.2011.10888.x.
- 25. Antic T, Hyjek EM, Taxy JB. The vanishing testis: a histomorphologic and clinical assessment. Am J Clin Pathol 2011; 136(6): 872 – 80.
- 26. Walsh TJ, Dall'Era MA, Croughan MS, Carroll PR, Turek PJ. Prepubertal orchiopexy for cryptorchidism may be associated with lower risk of testicular cancer. J Urol 2007; 178(4 Pt 1):1440-6.
- 27. Elder JS. Epididymal anomalies associated with hydrocele/hernia and cryptorchidism: implications regarding testicular descent. J Urol 1992;148(2 Pt 2):624-6.
- 28. Kraft KH, Mucksavage P, Canning DA, Snyder HM 3rd, Kolon TF. Histological findings in patients with cryptorchidism and testisepididymis nonfusion. J Urol 2011; 186(5): 2045 – 9.
- 29. McIntosh LA, Scrimgeour D, Youngson GG, Driver CP. The risk of failure after primary orchidopexy: an 18 year review. J Pediatr Urol 2013; 9(6 Pt A): 759 – 62.
- Stec AA, Tanaka ST, Adams MC, Pope JC 4th, Thomas JC, Brock JW 3rd. Orchiopexy for intraabdominal testes: factors predicting success. J Urol 2009; 182(4 Suppl): 1917 – 20.
- 31. Jeong SC, Lee S, Ku JY, Lee SD. Clinical characteristics and treatment of cryptorchidism in adults: a single center experience. World J Mens Health 2014; 32(2): 110-5.
- 32. Chung JM, Lee SD. Individualized Treatment Guidelines for Postpubertal Cryptorchidism. World J Mens Health 2015; 33(3):161 - 6