

CASE REPORTS

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Epispadias, a correctable cause of structural infertility in a middle-aged man: a case report

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Abstract

Background Epispadias is a rare form of genito-urinary congenital anomaly and presentation in the middle-ages is even rarer. The reproductive outcome of treatment which was almost immediate, highlights the life changing effect of appropriate treatment for this condition in a middle-aged patient. We hereby report this rare case of a middle-aged man presenting with complete male epispadias with background infertility and the outcome of effective treatment. (Reviewer #1. Comment 1).

Case presentation Our patient is a 54-year-old security guard who presented to our facility in February 2022 (Reviewer #1. Comment 3) with passage of urine from an abnormal opening since birth. Examination revealed a middle-aged man having ammoniacal smell. He had a spade like glans penis, ventral hooding, urethral meatus at the root of the penis dorsally with no chordee. He was diagnosed to have complete male epispadias (CME) for which he eventually had a modified Cantwell–Ransley procedure under general anaesthesia. About 3 months after the procedure, patient was able to impregnate his wife, a feat he will be achieving for the first time in his life.

Conclusions The right treatment irrespective of age at presentation could significantly improve the quality of life of patients as evidenced by our patient whom after surgery was able to impregnate his wife for the first time at 54 years of age.

Keywords Epispadias, Middle-aged, Modified Cantwell–Ransley, Reproductive outcome, Case report

1 Background

Complete male epispadias (CME) is a rare form of genito-urinary congenital anomaly which is usually detected at birth and treated in infancy and childhood periods. Presentation in adulthood is even rarer more so in the middle-ages. We present a middle-aged man who reported to our hospital for the first time in adulthood at the age of 54 years (the oldest ever reported according to our literature search). This was highly intriguing considering the striking nature of epispadias which makes it easily noticeable from birth. The reproductive outcome of treatment

and improvement in self-esteem which was almost immediate highlights the life changing effect of appropriate treatment for this condition in a middle-aged patient.

2 Case presentation

Our patient is a 54-year-old security guard who presented to our facility in February 2022 (Reviewer #1. Comment 3) with passage of urine from an abnormal opening since birth. Had associated history of urgency incontinence (Reviewer #1. Comment 6.) but no systemic symptoms. Has no co-morbidity and no family history of similar illness. In his second order of marriage of 6 years, having intercourse but has no child. His first marriage was at the age of about 20 years but the wife left him after a few months.

He was told his parents had presented to a peripheral hospital in a rural area in childhood where they were informed nothing could be done and subsequently, he had resigned himself to fate even after he was informed, he could get help. Patient did not seek for any medical

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intervention on his own volition prior to his presentation in our facility (Reviewer #1. Comment 4) He remained with his symptoms until staff at his employing company noticed the occasional urine like smell around him and urged him to present to the hospital. He declined presenting to the hospital several times (Reviewer #3. Comment 1) until eventually a colleague of his, offered to shoulder the financial burden.

Examination revealed a middle-aged man having ammoniacal smell. He had a spade like glans penis, ventral hooding, urethral meatus at the root of the penis dorsally with no chordee.

He had a micturating cysto-urethrogram done pre-operatively which revealed a good capacity bladder. Abdomino-pelvic ultrasound scan done revealed structurally normal internal organs. He also had laboratory investigations done which included haemogram, urine microscopy culture and sensitivity (Reviewer #1, Comment 7) and renal function tests which all turned out to be within normal limits. A diagnosis of complete male epispadias was then made.

The patient eventually had a modified Cantwell–Ransley procedure under general anaesthesia in June 2022. Surgery was led by the most senior and experienced urologist in our hospital with 12 years' experience as a consultant urologist. Intra-operatively, following adequate cleaning and draping done, a traction stitch was placed at the glans penis. This was followed by a strip incision at the lateral edges of the urethral plate and circumferentially at the level of the prepuce allowing for the penis to be degloved. The urethral plate which was wide enough as visualised in the pre-operative images was then separated from the underlying cavernosal structures and tubularised over a size 16 fr Foley silicon catheter using a continuous running suture (polyglactin 4/0). The glanular wing were then raised and approximated distally. The Corpora cavernosa were then rolled over the tubularised urethra and an anchoring suture between both corpora were placed at intervals to hold them in place dorsally. The overlying skin was then approximated with interrupted sutures using polyglactin 2/0 sutures. (Reviewer #1. Comment 5 and 9).

He had an uneventful recovery, commenced oral intake 24 h post-operatively following a delayed recovery from general anaesthesia (Reviewer #1. Comment 8) and had his urethral catheter removed on the 14th day post-operatively and discharged thereafter. About 3 months after the procedure, patient was able to impregnate his wife, a feat he will be achieving for the first time in his life. Patient has since been voiding via his neo-urethra since surgery without difficulty. At his follow-up visit post-operatively, he reported that his earlier complains of urgency incontinence had subsided. (Reviewer #3. Comment 2).

3 Discussion

Complete male epispadias (CME) is a rare congenital anomaly [1]. With no reliable data stating its prevalence in Africa, we however can deduct from a Europe-wide prevalence evaluation done in 2010 which estimated a prevalence for isolated epispadias to be about 1:96,800 [2].

CME is a strikingly obvious congenital anomaly often times corrected in childhood [3]. Presentation in adulthood is not so common with no known earlier reported case in a middle-aged patient. Our patient presented for the first time at the age of 54 years. The oldest earlier reported case from sub-Saharan Africa was at 25 years of age [4]. In the series reported by Spinoit et al. [1], the mean age for surgeries was 13 months while Nerliu et al. [5] reported a mean age of 9.13 years in their series, these are in sharp contrast with our index case. Epispadias is a known cause of low self-esteem amongst growing boys [4] and probably ignorance as highlighted in our case report following patients first presentation in childhood and also poverty considering the patient's willingness to finally present to the hospital after his colleagues offered to bear the cost may have accounted for the very late presentation of our patient.

The modified Cantwell–Ransley procedure has been reported across the world as a reliable repair technique with good functional and cosmetically acceptable outcome [3–5]. This was replicated in our patient where we equally recorded a good functional and cosmetic outcome post-operatively.

Sexual function has been found to be well preserved in patients with CME [6, 7]. Some have even found no difference in sexual function between those in the general population and their study participants [7]. Our index patient reported no problems with his sexual function in keeping with earlier findings. On the other hand, despite his age, our index patient was able to impregnate his wife after surgery in contrast with earlier reports of sub-fertility in such patients [7]. A number of other studies have also reported a low fertility rate of 33% either as a result of poor ejaculatory function or due to poor semen parameters [8, 9].

4 Conclusions

Though presenting very late in adulthood, this patient had a significant improvement in quality of life after surgery including his ability to impregnate his wife for the first time at 54 years of age after appropriate treatment. There is a need for more public health enlightenment in this part of the world to avoid such late presentation of patients in the future. (Reviewer #1. Comment 10) This further signifies the effectiveness of the modified Cantwell–Ransley procedure even when applied to middle-aged patients with CME as further expressed by the index patient who has expressed satisfaction with his treatment outcome.

Appendix 1

See Fig. 1.



Fig. 1 Pre-operative picture

Appendix 2

See Fig. 2.



Fig. 2 Op-site at 2 weeks post-operatively

Appendix 3

See Fig. 3.



Fig. 3 Patient micturating at 2 weeks post-operatively

Abbreviation

CME Complete male epispadias

Author contributions

All authors were involved in the pre-operative assessment of the patient. MK, KA and AA were involved in the surgery. All authors were involved in the post-operative management and follow-up of the patient.

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Availability of data and materials

Data as reported are available in the patient's hospital case notes.

Declarations

Ethics approval and consent to participate

Informed consent to participate was obtained from the patient after adequate counselling.

Consent for publication

Consent for publication was obtained from the patient after adequate counselling.

Competing interests

All authors declare no competing interests.

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