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- 1 Top 50 most cited articles on prostatic artery embolization for benign prostatic hyperplasia: A bibliometric review

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Top 50 most cited articles on prostatic artery embolization for benign prostatic hyperplasia: A bibliometric review

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Abstract

BACKGROUND

Bibliometric analysis can be used to assess the current state of the literature and publication trends on a given topic. There has not been a review of this kind on prostatic artery embolization (PAE) for benign prostatic hyperplasia (BPH). PAE is a relatively new and somewhat controversial treatment option for BPH. Given the novelty and controversy, there has been much research published on the topic recently.

AIM

To survey the current state of research on PAE for BPH by using bibliometric analysis to analyze the top 50 most highly cited articles.

METHODS

A cross-sectional study was performed using the Web of Science database to identify the most cited articles published on PAE for BPH as of June 2022. Articles that did not primarily focus on PAE or BPH as an indication were eliminated. The 50 most cited articles were carried forward for analysis.

RESULTS

All but 6 articles were published in the last decade with contributions from 15 countries. Fifty-two percent of the studies had a C level of evidence. The majority were published in the *Journal of Vascular and Interventional Radiology* and *Cardiovascular and Interventional Radiology*. Twenty percent ($n = 10$) of the articles were published in urologic journals. On average, articles published in urologic journals tended to be more recent. The mean year of publication for an article in a urological journal was 2016.6 compared to 2013.9 in a non-urologic journal ($P = 0.02$). Seventy percent of the articles focused on clinical outcomes, while only 2% focused on practice guidelines. Self-citations accounted for 11.4 citations per

article on average, corresponding to 14.7% of all citations analyzed.

CONCLUSION

The most influential papers on this topic represent a fairly recent body of work with contributions from a wide variety of countries and journals. The fact that articles in urologic journals were published significantly more recently than articles on the list in non-urologic journals may suggest that the field of urology is starting to accept PAE for a wider range of indications. Finally, while there has been much high-quality research published, more influential studies on practice guidelines and technique may be beneficial.

Key Words: Prostate artery embolization; Benign prostatic hyperplasia; Endovascular urology; Bibliometric review

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Core Tip: Prostatic artery embolization is a technique for treating certain patients with benign prostatic hyperplasia that although controversial is gaining traction. We utilized bibliometric review to investigate the current landscape of the literature on this topic. Based on the publication trends in the most influential papers on this topic it appears that urologists may be starting to recognize the merits of the procedure. Additionally, this procedure has an international appeal with literature being published out of a wide variety of countries in radiology and urology journals alike.

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INTRODUCTION

Prostatic artery embolization (PAE) for the treatment of benign prostatic hyperplasia (BPH) is a relatively new, interventional radiology (IR) procedure that may serve as an alternative to traditional surgical methods in some patients. Articles on PAE for BPH began to be published in the early 2000s and since then, it has become a prominent area of IR and urologic research[1]. PAE has been shown to be an effective procedure, with success rates at 1-3 years and > 3 years post-procedure of 81.9% and 76.3% respectively[2]. Some have cited possible benefits over traditional surgical methods including shorter hospital stay, lower rates of urinary incontinence, and lower rates of retrograde ejaculation[3,4]. Nevertheless, there remains controversy surrounding the use of PAE to treat BPH by some professional societies indicating that ongoing investigation is necessary[5].

Bibliometric studies have been heralded as a way of measuring that which is inherently difficult to quantify, the quality and scope of the body of research for a specific topic[6]. They have been used to identify trends, find gaps within the evidence base, and provide guidance into new areas of study[7]. Of the many types of bibliometric reviews, citation analysis has become a popular methodology for evaluating a given subject[6,8]. Citation reviews in particular allow researchers to identify hot or controversial topics within the field, determine the nations that are leading the field, and can quantify the strength of the current evidence overall. This type of citation analysis has not previously been done on PAE for BPH.

Citation analysis was used to identify the characteristics of the current state of the literature surrounding PAE for BPH. A search was conducted for the 50 most highly cited articles within the current body of evidence. Given the controversy surrounding the subject, care was taken to identify the level of evidence present within each article.

MATERIALS AND METHODS

In June 2022 the Clarivate Web of Science (WoS) database was used to identify articles that discussed PAE for BPH using the search terms: (PAE AND Prostate) OR [(Prostatic OR Prostate) AND (Embolization OR Embolisation)]. Fourth year medical students trained in bibliometric review performed the search, any discrepancies were discussed at length based on full text review. These articles were sorted by number of citations in descending order. Article titles and abstracts were used as

an initial screen for inclusion, which was performed by a single author. Duplicate articles and those not focusing on PAE for BPH were excluded. The remaining articles were exported for further full text analysis.

The WoS database extracted several variables of interest pertinent to each article, including title, journal, citation count, year published and country of origin. Other variables of interest, such as level of evidence and article type, were extracted through full-text review by reviewers. Self-citation search was carried out by authors using analysis of citations within Google Scholar. Variables were extracted from the WoS database and exported to Microsoft Excel which was used to store, analyze, and visualize the data. The level of evidence was determined independently by two authors using guidelines from the *Journal of Vascular and Interventional Radiology* 2018 methodology update and Oxford Centre for Evidence-based Medicine[9,10]. Prior to the database search, article categories were determined to consist of clinical outcomes, technique, imaging/anatomy, translational, guidelines, and other. Each article was classified into only one category based on the consensus opinion of two authors.

A *t*-test was used to compare the mean year of publication between articles on the list that were published in urology journals compared to those that were not. Statistical significance was determined as $P < 0.05$.

RESULTS

The initial search generated 851 articles. The titles and abstracts were reviewed for the first 120 articles. 58 were found not to focus on PAE specifically for BPH and were excluded from analysis leaving 62 articles that were exported for full text review. 2 additional articles were excluded after full text review, which determined that they were only tangentially related to PAE for BPH (Figure 1)[11,12]. Of the remaining articles, the 50 most cited were analyzed in this review (Supplementary Table 1).

All articles were published between 2000 and 2020 with 46 published between 2011 and 2020 (Figure 2). Additionally, 9 of the top 10 articles were published during this period. The average number of citations was 77.3 with a range of 34-171 and median of 64.5. Nine articles were considered neither positive nor negative towards PAE for BPH, of the remaining 41 articles analyzed 40 (97.6%) were considered to be positive in favor of PAE for BPH.

The top 50 most cited articles represent the work of researchers from 15 different countries. Brazil had the highest number of contributions ($n = 12$), followed by Portugal ($n = 9$), the United States ($n = 8$), and China ($n = 5$) (Figure 3).

The analyzed articles were published in 13 different journals with over half published in the *Journal of Vascular and Interventional Radiology* ($n = 15$) and *Cardiovascular and Interventional Radiology* ($n = 15$). *Radiology* was the next most productive journal ($n = 5$) with all other journals having fewer than 4 contributions (Figure 4).

The most common level of evidence was C ($n = 26$), followed by B ($n = 8$), D ($n = 7$), E ($n = 6$), and A ($n = 3$) (Figure 5). The majority of articles were classified as clinical outcomes ($n = 35$), followed by technique ($n = 6$), imaging/anatomy ($n = 5$), translational ($n = 2$), guidelines ($n = 1$), and basic science ($n = 1$) (Figure 6).

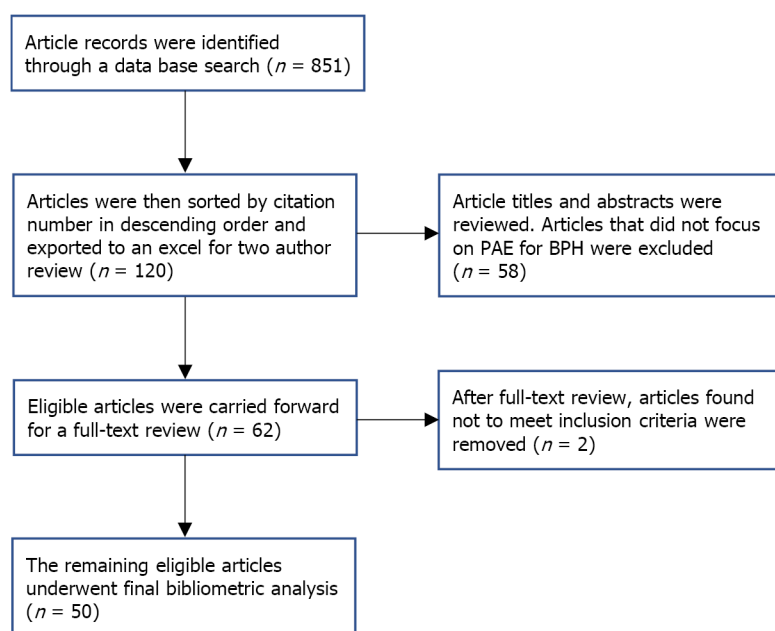
Twenty percent ($n = 10$) of the articles were published in urologic journals. On average, articles published in urologic journals tended to be more recent. The mean year of publication for an article in a urological journal was 2016.6 compared to 2013.9 in a non-urologic journal ($P = 0.02$).

Self-citations accounted for 14.7% of all citations, or an average of 11.4 self-citations per article, for the top 50 most cited articles. Seventy-two percent of the articles analyzed had greater than 10% of their citations coming from the authors of the article themselves. Two articles had no self-citations.

DISCUSSION

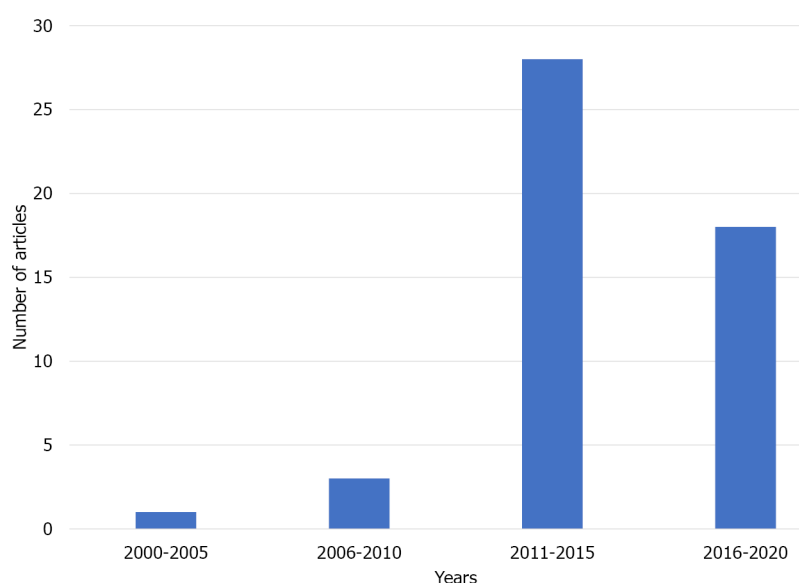
The most cited studies of PAE for BPH were published out of a variety of countries, with Brazil, Portugal, the United States, and China being particularly well represented. Some of the first research on PAE was conducted in China, so it is unsurprising that the number one cited paper was published by a research group based there. Additionally, much of the early research by Wang is represented at various positions in the list, including at the 21st, 33rd, and 36th spots.

Though articles on the topic were first published in the early 2000s, many of the most highly cited articles have been published in the last decade. This illustrates both continued high-quality research and the growing popularity of the procedure based on a track record of safety and efficacy. Many of these recent articles focus on clinical outcomes. This is not surprising given surgical therapies such as TURP have long been accepted as reliable options for the treatment of BPH refractory to medical management. Surgical management has well demonstrated benefits and clearly outlined risks, therefore much work had to be done to demonstrate the noninferiority of PAE as it became more commonplace. The Society of Interventional Radiology, along with other professional societies have continued to support this kind of research, urging for larger trials with longer follow-up periods to compare PAE to established



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Figure 1 Modified PRISMA flowchart. PAE: Prostatic artery embolization; BPH: Benign prostatic hyperplasia.



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Figure 2 Frequency of top 50 articles published between 2000 and 2020.

surgical therapies[13].

It is likely that strong research in the field has been important for the adoption of PAE as an established therapy by practice groups and specialty societies. Additionally, with increasing interest and access to scientific literature, this research is also important in informing patients who may be interested in a variety of treatment options for BPH. As the efficacy and safety of PAE continues to be demonstrated, the proportion of patients who self-refer for the procedure may also be growing[14]. Despite this body of research, PAE is not a universally recognized alternative to existing therapies in appropriate patients[12,15]. Though outcomes research will likely remain a popular article subject, over time it is possible that more of the influential research in the field will shift to updating practice guidelines and improving techniques.

Despite strong resistance early on, our findings suggest that the field of urology may be starting to accept PAE for a wider range of indications. This is supported by the fact that articles in urologic journals were published significantly more recently than articles on the list in non-urologic journals.



Figure 3 Frequency of articles published by country. PAE: Prostatic artery embolization; UK: United Kingdom; USA: United States.

Still, official urologic guidelines such as those published by the American Urologic Association still do not support PAE for BPH[12]. Continued publication of high impact research on this topic may indicate slowly changing opinions on these specific indications in the field.

The majority of the top articles had a “C” level of evidence, which was somewhat expected given the novelty of the field. Performing a randomized controlled trial (RCT) that meets the criteria for an “A” level of evidence is challenging in any field, however some of these barriers are more apparent when investigating PAE. Given that PAE is a relatively new treatment option, patient volume can be a concern when attempting to construct large scale RCTs. The case volume at some centers is growing, which will likely enable larger scale RCTs as well as cohorts for retrospective analysis[14].

Though high quality RCTs are considered the gold standard, not all clinical questions lend themselves to this type of analysis, and the value of other well-designed studies should not be discounted. The utility of observational studies in complementing randomized trial findings is clearly demonstrated by the United Kingdom ROPE study, which showed PAE could be a cost-effective surgery alternative in certain healthcare systems[3]. The ease of performing these large-scale studies will only improve with increased procedure volume. Increased recognition by patients and specialty societies will also aid in opening the door for more high-level research.

The highest cited paper of the group, “Benign Prostatic Hyperplasia: Prostatic Arterial Embolization vs Transurethral Resection of the Prostate-A Prospective, Randomized, and Controlled Clinical Trial” by Gao *et al* [16] garnered 171 citations and was one of a handful to earn an “A” level of evidence. This paper was the culmination of several smaller studies by Gao *et al*[16] and the rest of his China-based team, some of which represent the earliest PAE related research found[1]. This is a high quality, well designed RCT that came at a time when there were few prospective randomized studies within the literature. Despite its relatively small sample size, it was a critical early study in affirming the efficacy of PAE for BPH, even when compared to the gold standard operative management. It also was among the first to outline a specific methodology for performing PAE, which others continue to build on. As the first RCT on the subject, its position at the top of the list is not surprising.

“Embolization of prostatic arteries as treatment of moderate to severe lower urinary symptoms (LUTS) secondary to benign hyperplasia: results of short- and mid-term follow up” by Pisco *et al*[17] was cited 155 times, the second most cited article of those evaluated. This early, large, and well-designed prospective study demonstrated the safety and efficacy of PAE in treating moderate and severe, medically refractory BPH. Having been published in 2013, this study came at a time when there were few nonsurgical options for these patients. Contributing to the popularity of this article is the framework the study established, one that other authors such as Carnevale *et al*[4] would use in later prospective randomized

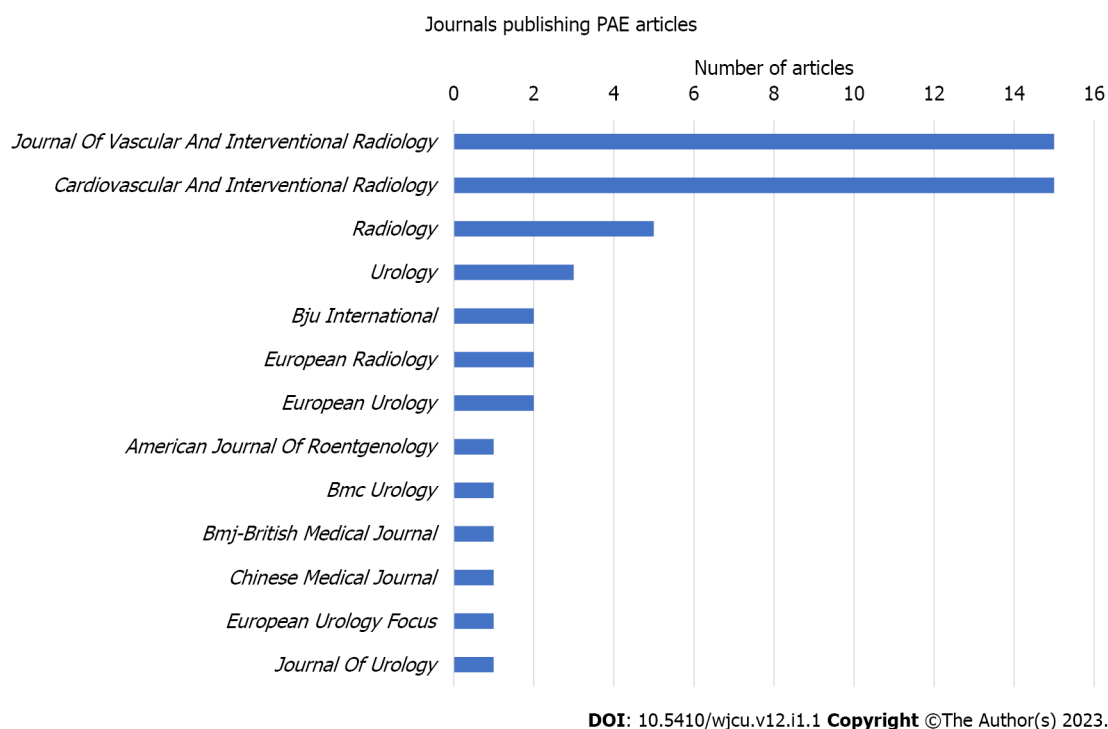


Figure 4 Frequency of articles published by journal. PAE: Prostatic artery embolization.

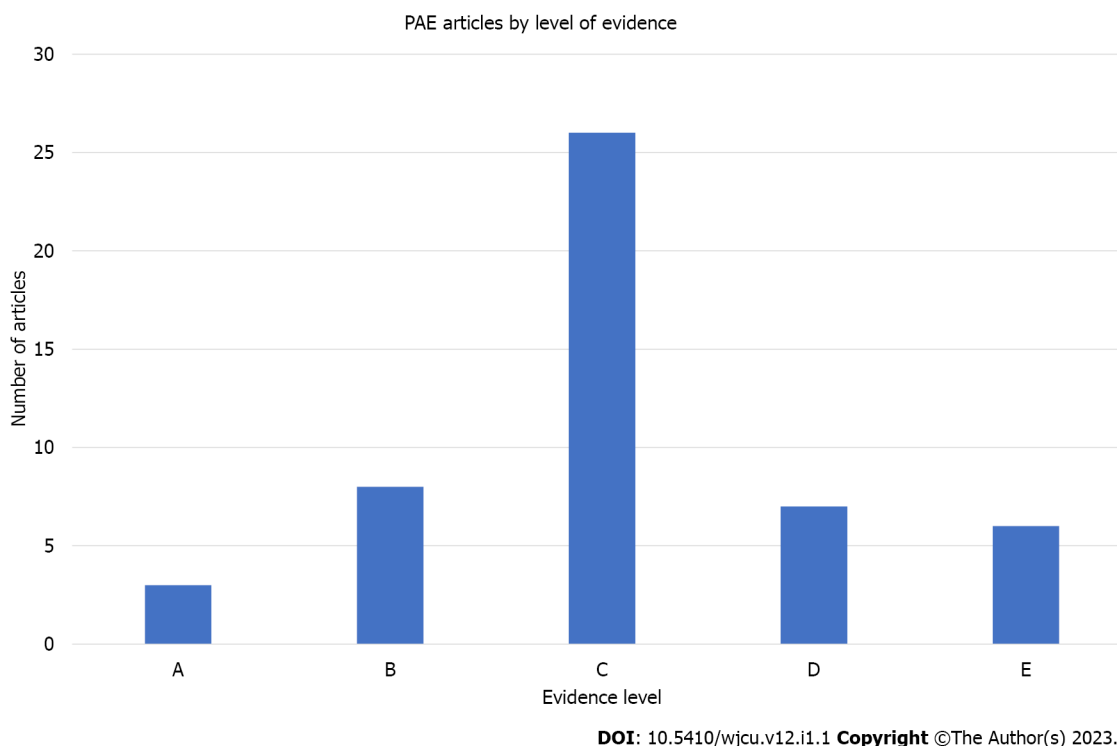
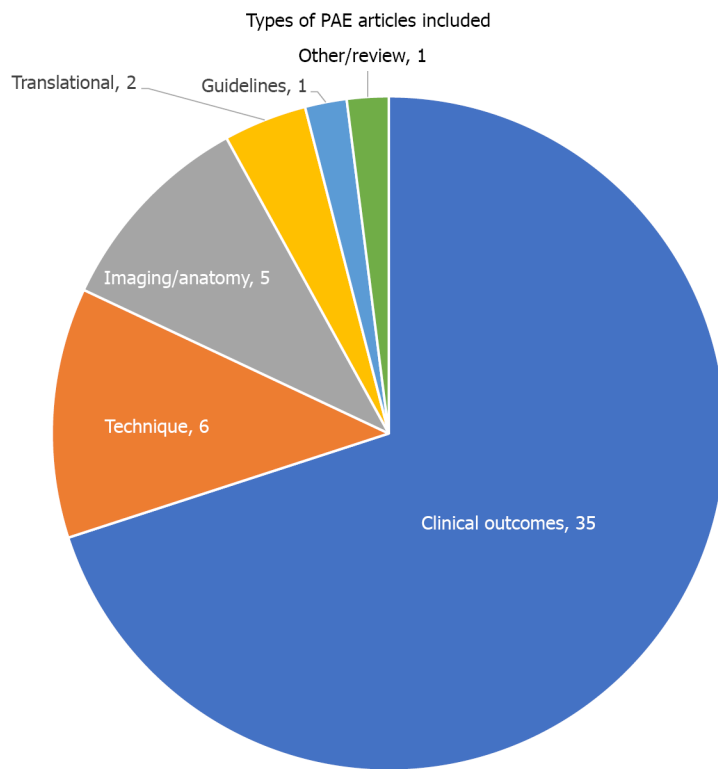


Figure 5 Frequency of articles published by level of evidence. PAE: Prostatic artery embolization.

trials.

With 134 citations, the third most highly cited article on this list is 2015's "Transurethral Resection of the Prostate (TURP) Versus original and PErFecTED Prostate Artery Embolization (PAE) Due to Benign Prostatic Hyperplasia (BPH): Preliminary Results of a Single Center, Prospective, Urodynamic-Controlled Analysis" by Carnevale *et al*[4]. Only the second RCT published on PAE, it sought to expand upon the works published by Gao *et al*[16] and Pisco *et al*[17]. This study utilized urodynamic evaluation to assess patient's bladder dysfunction, something previous studies had not evaluated, likely due to the



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Figure 6 Frequency of articles published by article type. PAE: Prostatic artery embolization.

invasiveness of the testing. The use of these established metrics for evaluating participants was important in producing a more quantitative comparison between PAE and TURP. Additionally, this study included a third arm allowing for the evaluation of two different methods for PAE, something not done before. The study's findings supported earlier works by other authors and laid the foundation for future PAE research.

At first glance, the self-citation rate of 14.7% may seem unreasonably high, leading to questions of the validity of the research of PAE for BPH. This is explained, however, by the status of many of these papers as landmark studies within the PAE literature. In this setting, the high self-citation rate corresponds to the continued work of many prolific authors, whose citing of themselves is quite reasonable as they are often the innovators who created and published on many of the popular techniques employed today.

The goal of the bibliometric review is to analyze the most influential articles in a given field. Despite contributing to the literature on this topic, the analysis does have some limitations. Citations were the only measure used within this study to rank articles. Although considered a good measure, citations do not consider all dimensions of influence an article may have. This measure is also biased towards older articles which have had more opportunity to be cited. Newer, potentially more influential articles may not have been included due to inadequate time to accrue citations. Citation counts for each article were generated by WoS which does not count citations from every journal. Finally, citations in certain types of publications such as books, patents, conference proceedings, and dissertations, among others, are not counted. As a result, the full influence of some publications may not be fully reflected by the analysis.

CONCLUSION

The most influential research articles on PAE come from a variety of countries and represent a fairly recent body of work with all but 6 published in the last decade. The majority of these highly cited works were published in either JVIR or CVIR, though 20% of the articles were published in urologic journals. Both of these fields will likely remain important sources of influential research on this topic. Though PAE is more widely accepted today, some still feel there is insufficient evidence to support it for BPH. The sustained resistance illustrates the continued need for large, high-quality studies to corroborate previously published findings, outline improved techniques, and develop impactful and up-to-date practice guidelines to ensure optimal patient outcomes.

ARTICLE HIGHLIGHTS

Research background

Prostate artery embolization (PAE) for benign prostatic hyperplasia (BPH) is a new technique hardly past its infancy. Past studies have evaluated the utility, indications, efficacy and basic science of the procedure. Although there have been some systematic studies, a bibliometric review has not been conducted prior to this study.

Research motivation

This bibliometric review will analyze the 50 most cited articles with regards to PAE for BPH. This will show the strengths and weaknesses of the current body of literature, where said literature is being produced, and will attempt to quantify the growing acceptance of this procedure by the field of urology.

Research objectives

By performing a bibliometric review, it is hoped that avenues for future expansion of the literature are elucidated. This will allow future researchers to better pursue novel and helpful research, rather than studies similar to those that have already been conducted and are well-respected.

Research methods

The bibliometric review, while popular in many fields, has not seen the same level of acceptance in the fields of interventional radiology or urology. As described previously, PAE for BPH is relatively cutting-edge and the literature on the subject is not yet well-established. Therefore, it is unsurprising that this study represents the first bibliometric analysis of PAE for BPH.

Research results

The top 50 articles on PAE for BPH were all written within the last two decades, with a heavy bias towards the last decade. Although only 20% of these articles were published in urology journals, those that were published within these journals were significantly newer than their non-urologically published counterparts. The C level of evidence was the most common level represented. Future research should include higher evidence level studies (comparative randomized control trials and meta-analyses) and may be more likely to be accepted into urology journals.

Research conclusions

The methodology of applying a bibliometric review to PAE for BPH is a completely novel endeavor. This theory proposes the idea that the literature for PAE for BPH is young but growing. Additionally, our results suggest that PAE for BPH is steadily gaining greater acceptance within the field of urology.

Research perspectives

Future research should be directed towards high evidence level studies. This will steadily increase the evidence level seen by future bibliometric reviews as would be expected in a young but rapidly expanding field.

FOOTNOTES

Author contributions: Zoppo CT contributed to conceptualization, methodology, and writing; Taros T contributed to data curation, methodology, and writing; Harman A contributed to supervision, reviewing, and editing.

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CASE REPORT

- 10 Phallic rubber band application to prevent enuresis unusual cause of urethral stricture in a child: A case report

Khalid A, Nasiru M, Abdulwahab-Ahmed A, Muhammad AS, Agwu NP, Lukong CS

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Phallic rubber band application to prevent enuresis unusual cause of urethral stricture in a child: A case report

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Abstract

BACKGROUND

Urethral stricture is the abnormal narrowing of the urethra due to spongiofibrosis. The established aetiological factors for urethral stricture abound in our environment. However, the application of a penile rubber band to prevent enuresis thereby causing this pathology is not a familiar occurrence. Patients with enuresis can suffer psycho-social challenges and trauma, especially for a child due to peer stigmatization. This has a great impact on the child's psyche and may affect even his performance at school. The aim of this paper is to highlight the psychosocial impact and management challenges of a child with enuresis (nocturnal urinary incontinence).

CASE SUMMARY

This is the case of a 10-year-old boy who presented with a history of nocturnal urinary incontinence since birth and lower urinary tract symptoms (LUTS) for 3 years culminating in chronic urinary retention. He maintained a normal urge to micturate and continent of urine during the daytime before the onset of LUTS. He had an antecedent longstanding history of tying a rubber band to the penile shaft mostly before going to the bed at night to prevent enuresis due to his peer stigmatization. He was acutely ill-looking, with distended suprapubic region. The phallus and scrotum were enlarged and oedematous with a circumferential proximal penile shaft scar and ventral penile shaft urethrocutaneous fistula. He was diagnosed to have complete short segment bulbopenile stricture and right ectopic ureter. He subsequent had augmented anastomotic urethroplasty and

bilateral non-refluxing ureteroneocystostomy at different sitting.

CONCLUSION

The adherence to surgical principles of urethral stricture and enuresis management where surgically correctable in a child is associated with the resolution of enuresis and social reintegration.

Key Words: Enuresis; Double ureter; Ectopic ureter; Rubber band; Urethral stricture; Case report

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Core Tip: Enuresis is a psycho-socially challenging and emotionally traumatic condition. This paper brings to light the management of secondary nocturnal enuresis in a 10-year-old male child with an attendant urethral stricture. A complication resulting from the longstanding application of rubber band to the penile shaft to prevent enuresis and stigmatization by his peers. The clinical presentation of this case is clouded by the clinical features of the dual pathology. Hence, a step-wise application of the surgical principle of clinical evaluation, laboratory and imaging investigations and surgical treatment is associated with resolution of enuresis and patient satisfaction.

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INTRODUCTION

Urethra stricture disease is a very common urologic condition in our environment[1]. It is the abnormal narrowing of the urethra due to spongiofibrosis[2]. It is the fourth most commonly diagnosed annual new urologic disease seen in our clinical setting among male adults[3]. Post-inflammatory aetiology mainly from post-gonococcal urethritis closely followed by posttraumatic causes due to road traffic accidents or falling astride is encountered frequently[1,3]. In our setting, urethral stricture disease is a less common condition among the paediatric population when compared to the adult population. Urethral stricture when seen in children is often idiopathic or as an iatrogenic consequence commonly resulting from urethral catheterization or hypospadias surgery, especially in younger children usually involving the anterior urethra[4]. In older children, traumatic aetiologies from pelvic injury predominate and commonly affect the posterior urethra[4]. Other reported causes though rare in older children include post-infective urethritis and from lichen sclerosis[4]. The phenomenon of phallic strangulation by a wide variety of objects ranging from nonmetallic to metallic objects has been reported commonly among adults that may result in urethral stricture though rarely encountered in children[5]. Several reasons adduced for this include presence of psychiatric conditions, induction or enhancement of erection for those with erectile dysfunction, and avoidance of coitus were advanced to explain why adults strangulate phallus[5]. In children, however, the actual reason may differ from adults and attempt at controlling enuresis may be a possible reason. The peer ridicule, stigmatization influence may be a strong motivator in children. The application of phallic rubber to prevent enuresis is not a familiar practice among children with enuresis in our environment, let alone urethral stricture as its complication. The application of constricting devices particularly when prolonged initially causes phallic tissue ischemia, urethral erosion and fistulation. If complicated by urinary tract infection or urethritis, the subsequent healing by fibrosis may results in urethra stricture formation. The key investigative modalities for urethral stricture diagnosis according to American Urological Association (AUA) guidelines are urethrocystoscopy, retrograde urethrography, voiding cystourethrography, or ultrasound urethrography[6]. The management of urethra stricture should be guided by the strict application of its surgical principles.

Enuresis is a psychosocially challenging and emotionally traumatic condition. According to the American Pediatric Academy, it refers to the involuntary wetting of clothes or bedding by urine during the daytime or nighttime for a period of at least 3 consecutive months in children older than 5 years of age[7,8]. The nighttime bed wetting or nocturnal enuresis occurs more frequently and tends to be multifactorial, and affects mostly girls[8]. Family history is a strong risk factor[8]. It can be primary, being the most common type in which there is no period of 6 consecutive months of nighttime urinary control[8]. The secondary type of enuresis is associated with organic or psychological aetiologies and in this variety; the children have a periods of 6 consecutive months of nighttime urinary control before the onset of incontinence [8]. In girls primary enuresis is a common occurrence while secondary enuresis predominates in boy with ectopic ureter being a culprit[8-10]. The management of enuresis involves clinical evaluation, relevant laboratory investigations such as urinalysis and urine culture, and urinary tract imaging investigations: Namely ultrasound scan, radiography, computerized tomography scanning, and magnetic resonance imaging as may be necessary. Its treatment ranges from nonsurgical and nonpharmacological options such as urotherapy, limitation of fluid intake, and bedwetting alarm to pharmacological agents such as desmopressin, imipramine, and Oxybutynin among other agents and rarely surgery where a definite amenable cause is found[8]. Other treatment options worth mentioning are Transcutaneous Parasacral

Electrical Nerve Stimulation and Functional Magnetic Stimulation[8].

The clinical entity of urethra stricture arising from a rare aetiology such as phallic rubber band application as an attempt at self-treatment of enuresis in a child is scarce in the literature. To the best of our knowledge, this is the first reported case of its kind. We report this unusual cause of urethral stricture in a male child with enuresis and its holistic management. The aim of this paper is to highlight the psychosocial impact and management challenges of a child with enuresis (nocturnal urinary incontinence).

CASE PRESENTATION

Chief complaints

This is the case of a 10-year-old boy who presented with a history of nocturnal urinary incontinence since birth and lower urinary tract symptoms (LUTS) for 3 years.

History of present illness

He was noticed by parent to be having nocturnal urinary incontinence beyond the age of achieving urinary continence. Three years before presentation, he had voiding and storage LUTS associated with leakage of urine from the ventral surface of the penile shaft. The symptoms progressively worsened over time culminating in chronic urinary retention. Before onset of LUTS, he maintained a normal urge to micturate and the continence of urine during the daytime. The attempt at urethral catheterization at presentation to our facility was unsuccessful warranting open suprapubic cystostomy. There was an associated history of recurrent fever, anorexia, nausea, and penile swelling for which he was treated on several occasions for urinary tract infections at the referral health facility. He had an antecedent longstanding history of tying a rubber band to the penile shaft mostly before going to the bed at night to prevent enuresis due to his peer stigmatization (Figure 1A and B).

History of past illness

There was no remarkable past medical or surgical history.

Personal and family history

There was no associated history of psychiatric illnesses, family history of enuresis, or other remarkable histories.

Physical examination

He was acutely ill-looking, not in painful or respiratory distress, afebrile, pale, and dehydrated but not jaundiced with ammoniacal odour. His temperature, respiratory rate, pulse rate, and blood pressure were 36.1 °C, 24 cycles/min, 102 beats/min, and 100/60 mmHg respectively. The abdomen was full and moved with respiration. The suprapubic region was distended and stony dull to percussion. The phallus and scrotum were enlarged and oedematous with a circumferential proximal penile shaft scar and ventral penile shaft urethrocutaneous fistulous exit at the scar site (Figure 1B). The digital rectal and other systemic examinations were unremarkable.

Laboratory examinations

The complete blood cell count was $7.4 \times 10^9/L$ and pretransfusion and posttransfusion packed cell volumes of 24% and 30.6% respectively. The serum urea was 18.1 mmol/L, creatinine 2.7 mg/dL, and electrolytes: Na^+ 132 mmol/L, K^+ 3.6 mmol/L, HCO_3^- 18 mmol/L and Cl^- 96 mmol/L. The urinalysis was positive for nitrites, leukocytes, blood, and protein. The urine culture yielded growth of *Staphylococcus aureus* sensitive to cefotaxime among others.

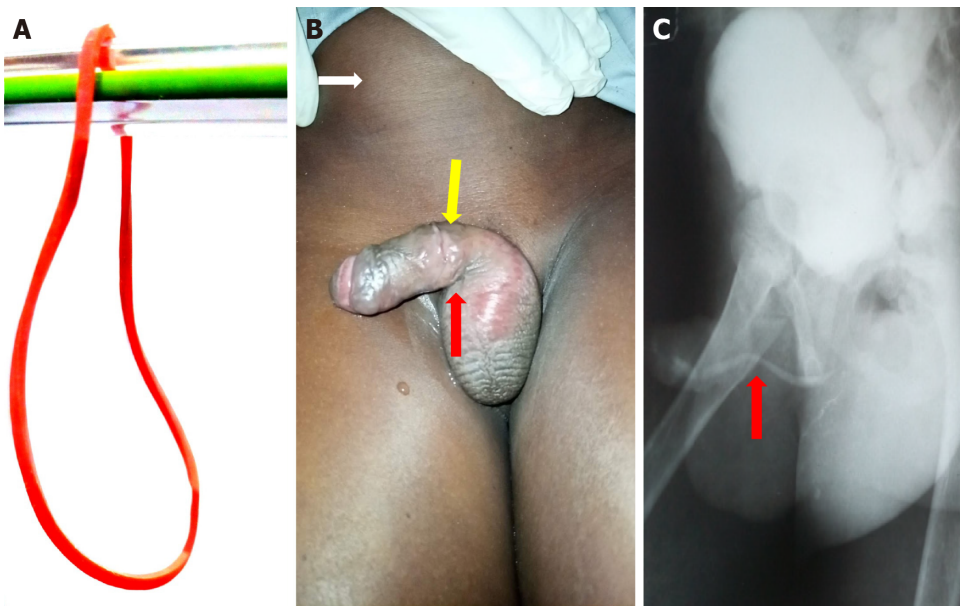
Imaging examinations

Abdominal Ultrasound scan showed a grossly distended bladder with bilateral moderate to severe hydronephrosis with bilateral hydroureters. The combined retrograde urethrogram and voiding cystourethrogram done later showed normal anterior and posterior urethra with an area of stricture in evolution but the contrast was noticed to be exiting the phallus ventrally at the site of application of the rubber band (Figure 1B and C), dilated bladder and left duplex system with grade IV vesicoureteral reflux into the duplex system (Figure 2A). There was no demonstrable reflux or visualization of the right ureter (Figure 2A).

The abdominal computerized tomography scan revealed bilaterally functioning kidneys with a partial and complete duplex (double ureters and collecting system) on the right and left respectively (Figure 2B-D), and the right lower ureter insertion into the region of the prostatic urethra and left duplex ureters insertion into the bladder (Figure 3A and B).

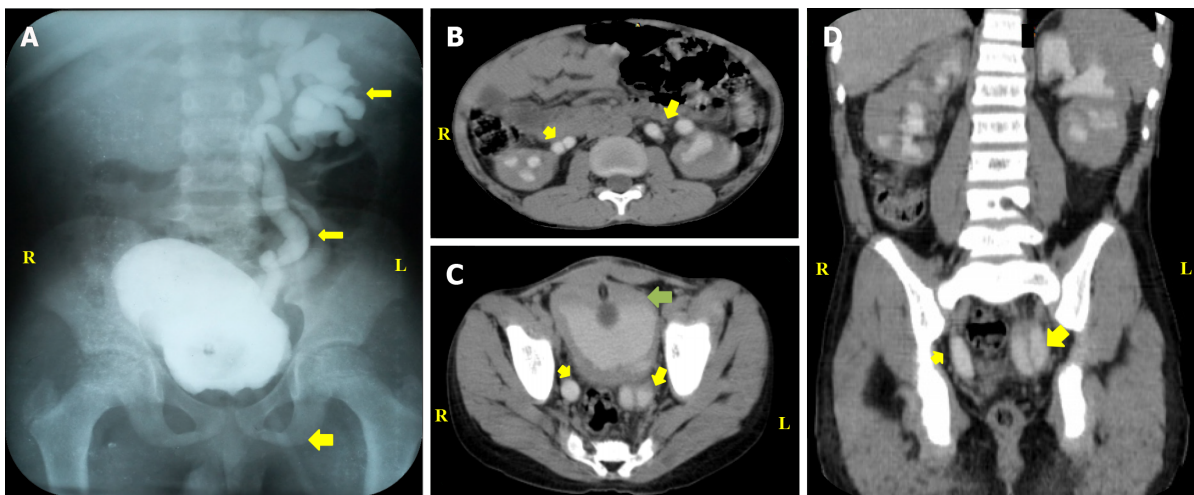
FINAL DIAGNOSIS

At this point, a diagnosis of enuresis secondary to the right ectopic ureter was made (Figure 3A) with a grade IV left duplex or double ureter reflux (Figure 3B).



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Figure 1 Initial patient presentation at the facility and follow-up contrast imaging. A: Showing typical elastic rubber band; B: Physical examination findings showing the distended suprapubic region from urine retention (white arrow), circumferential penile shaft scar-site of rubber band application (yellow arrow), and ventral urethrocuteaneous fistulous exit (red arrow); C: The combined retrograde urethrogram and voiding cystourethrogram showing the location of an evolving urethral stricture (red arrow).



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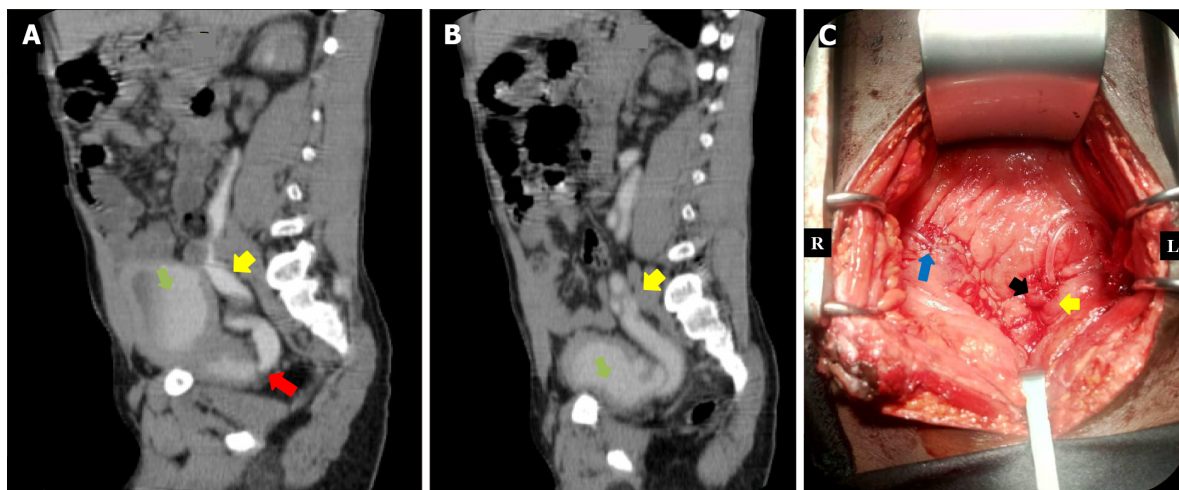
Figure 2 Contrast imaging studies. A: Static cystogram showing duplex left collecting systems and ureters with reflux (yellow arrows) and suprapubic catheter (broader yellow arrow); B: Computerized tomography scan, axial view showing bilateral duplex upper ureters (yellow arrows); C: Computerized tomography scan, axial view demonstrating right partial duplex and left complete duplex lower ureters (yellow arrows), and bladder (green arrow); D: Computerized tomography scan, coronal view of the ureters (yellow arrows).

TREATMENT

He had intravenous fluid, parenteral antibiotics, and a unit of blood transfusion. An initial suprapubic cystostomy was performed which drained urine of 1000 mL. He was discharged home on continuous bladder drainage and monthly suprapubic catheter (14fr) replacement until 8-mo later when Augmented Anastomotic Urethroplasty was performed. The intraoperative findings were a complete short-segment bulbopenile urethral stricture about 1.5 cm long with a fistulous tract arising adjacent to the proximal part of the stricture and ending blindly into the ventral penile shaft skin.

Thereafter, the caregiver was counseled and educated on conservative management of enuresis essentially involving lifestyle modifications with some improvement. However, enuresis persisted despite these measures leading to further imaging investigation (Figure 3A and B).

The parents were counseled and he had exploratory laparotomy with non-refluxing right ureteroneocystostomy and non-refluxing left duplex system ureteroneocystostomy (as common sheath reimplantation) with improvised (feeding



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Figure 3 Contrast imaging and intraoperative image. Sagittal views of computerized tomography scan and ureteroneocystostomy. A: Computerized tomography scan, sagittal view showing right ureter (yellow arrow) insertion into the region of prostatic urethra (red arrow) and bladder (green arrow); B: Computerized tomography scan, sagittal view demonstrating left duplex ureter (yellow arrow) insertion into the bladder; C: Intraoperative view of right ureteroneocystostomy (blue arrow) and left duplex ureteroneocystostomy (black and yellow arrows).

tubes) as external ureteric stents, **Figure 3C**. He had silicon urethral catheter drainage for three weeks. Both stents and catheter were removed at 3-wk postoperative day. He did well postoperative and was discharged home on the 14th postoperative day.

OUTCOME AND FOLLOW-UP

He was 2 years on the follow-up visits after last surgery; the enuresis stopped, and was well-integrated among his peers.

DISCUSSION

This case report brings to light an uncommon aetiology of urethral stricture in a child with enuresis. The clinical presentation of this dual pathology in a child presents the clinician with a herculean task due to the clouded picture of symptoms. In the index patient, although the history of nocturnal enuresis dated to birth, the thought of an organic cause was not clear at the time of presentation. Therefore, the application of surgical principles in the management of this dual pathology comes in handy. The urethral stricture was diagnosed using the combination of retrograde urethrogram and voiding cystourethrogram, and had urethroplasty, one of the diagnostic investigations and treatments respectively recommended by AUA guidelines[6]. After this treatment, he had the non-pharmacological treatment of enuresis including urotherapy and limitation of fluid intake, which is often the first line of treatment with good outcome in the majority of patients with primary nocturnal enuresis[8]. He had some symptomatic improvement. However, the improvement was short-lived as symptoms persisted later. This led to the reconsideration of our initial thoughts of primary nocturnal enuresis.

At this point, it became pertinent to consider secondary nocturnal enuresis in which organic or psychological aetiologies may be present[8]. Moreso, primary nocturnal enuresis is commonly seen in girls and family history is a strong factor[8]. The index patient is a male child and had no family history of nocturnal enuresis. Also during evaluation for the urethra stricture, the findings of the left refluxing duplex ureter a congenital anomaly, and non-visualization or reflux in the right ureter (**Figure 2A**) in a patient with infravesical obstruction is not consistent with the commonly observed bilateral vesicoureteral reflux and thus raised our suspicion of a possible congenital etiology or secondary nocturnal enuresis. This informed the decision to request for computerized tomography scan of the abdomen which confirmed the diagnosis of the right ectopic ureter and left duplex ureter (**Figure 3A and B**).

The right ectopic ureter is inserted into the region of the prostatic urethra (**Figure 3A**) bypassing the internal sphincter mechanisms, one of the male continence factors. This means the urine from the right kidney can only be controlled by the voluntary external sphincter. As such, in the daytime when the child is awake, he can maintain continence throughout the day but when deeply asleep at night he loses the voluntary external sphincter urine control resulting in secondary nocturnal enuresis. However, the patient maintained the urge to micturate despite enuresis because the duplex left ureter was inserted normally into the bladder (**Figure 3B**). Hence, urine from the left kidney enters the bladder under both internal and external sphincteric control. It is noteworthy that up to half the cases of male ectopic ureters inserts into the posterior urethra as observed in the index case aside from other rare locations[9]. As such urinary incontinence due to ectopic ureter is an uncommon presentation in male as observed in this case due the protective role of the external

sphincter. Conversely, female ectopic ureter inserts often into the female genital organs such as vagina which makes incontinence a prominent feature[10].

The index patient was managed based on surgical principles. This involves initial management of the urethra stricture followed by the nocturnal enuresis. Hence the initial emphasis here was to divert the urinary retention to aid control of infection and preservation of renal function. The patient presented with clinical and laboratory features of infection and renal impairments. Thereafter, he was investigated for the cause of urinary retention before the definitive urethroplasty for the complete short segment bulbopenile urethral stricture. Then, the parent was counseled on the nonpharmacological modality for the treatment of enuresis which led to a short-lived symptomatic improvement. The persistence of enuresis led to further investigation that confirms the diagnosis of the right ectopic ureter and left duplex ureter warranting bilateral non-refluxing ureteroneocystostomy (Figure 3C). This surgical treatment resolved the secondary nocturnal enuresis. The surgery has dual benefits. Firstly, it corrected the secondary nocturnal enuresis caused by the right ectopic ureter and secondly prevented the reflux of the left duplex system that is capable of compromising the renal function in the long term from recurrent urinary tract infection. Apart from open surgical treatment offered in this case, report of laparoscopic treatment amongst other operative techniques for managing patient with duplex ureter are available in the literature[11].

CONCLUSION

This paper aims to highlight the psychosocial impact and management challenges of a child with enuresis (nocturnal urinary incontinence). Urethral stricture following applications of the rubber band to the phallus is an uncommon presentation in children. Once urethral stricture is established, management should be guided by general principles of management of urethral stricture resulting from other aetiologies. Where failed conservative treatment of primary nocturnal enuresis occurs, secondary nocturnal enuresis should be considered and further investigations should be tailored toward unraveling the aetiology. The surgical treatment of enuresis is rewarding where the aetiology is known and associated with the complete resolution of the enuresis and social reintegration of the patient. This case management was challenging and we recommend the early use of imaging investigations in the evaluation of enuresis where symptoms persist despite conservative treatment.

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FOOTNOTES

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