Journal of Pediatric Urology (2015) 11, 85.e1–85.e11

A comparative bibliometric analysis of the top 150 cited papers in hypospadiology (1945–2013)

F. O'Kelly [a], G.J. Nason [b], L.C. McLoughlin [a], H.D. Flood [b], J.A. Thornhill [a]

[a]Department of Urological Surgery, Tallaght Hospital, Dublin, 24, Ireland

[b]Department of Urological Surgery, University Hospital, Limerick, Ireland

Correspondence to: F. O'Kelly, Department of Urological Surgery, Tallaght Hospital, Dublin, 24, Ireland, Tel.: +353 1 4414127; fax: +353 1 4412596

fokelly@rcsi.ie (F. O'Kelly)
nasong@tcd.ie (G.J. Nason)
louisemcloughlin@rcsi.ie (L.C. McLoughlin)
hugh.flood@hse.ie (H.D. Flood)
jturology@gmail.com (J.A. Thornhill)

Keywords
Hypospadias; Bibliometric analysis; Citation; Impact factor; Paediatric urology; Journal Citation Reports

Received 18 March 2014
Accepted 20 November 2014
Available online 4 March 2015

Summary

Introduction
Hypospadias, the most common congenital malformation of the penis, is characterized by an abnormal ventral opening of the urethral meatus, abnormal ventral curvature of the penis, and an abnormal distribution of the foreskin around the glans, with a ventrally-deficient hooded foreskin, and has been described in modern peer-reviewed literature since Noble's description in 1853. A dramatic increase in the literature on hypospadiology has focused on two main topics: The aetiology of the condition including molecular mechanisms and environmental influences, as well as surgical techniques to repair the defect, however there have been a number of facets of research stemming from this poorly understood condition in a developing subspecialty (Figure).

The majority of these publications focus on the evolution of surgical technique and management. Urethral reconstruction should offer the ability to stand to urinate and combine a satisfactory cosmetic result. The tubularized incised plate repair, popularized by Snodgrass et al., and the Mathieu repair, have been the mainstay for distal hypospadias, however, there have still been a large number of well-described techniques, with no consensus as to the ideal method of repair, some of which is attributed to non-uniform standards of reporting outcomes. There is also no standard objective means to qualitatively assess the importance of each of these contributions.

Objective
The objective of this study was to determine the top 150 peer-reviewed articles in the field of hypospadiology by way of citation analysis as a means to determine the importance/relevance of each contribution throughout 8 decades of research.

Study design
A cited reference search was carried out for indexed citations within Web of Science™ Core Collection. The top 150 indexed cited articles containing the topic "hypospadias" in specific subject categories were compiled using a component of Science Citation Index command. References were then evaluated, and analysed across a number of indices, and over time.

Results
The mean number of citations was 76 for each article. The top 150 articles were published across 21 countries, with the majority originating from the USA. The lead research institution was the University of Texas (Southwestern) Medical Center. In total, the top 3 centers represented 33 publications within the top 150 citations. The most commonly utilized journal was the Journal of Urology. The most cited author was W.T. Snodgrass. 39% papers in the top 150 cited articles were published prior to 1990. Three out of the five papers cited in the 1940s dealt with operative technique to repair hypospadias. The period 2000–2010 in contrast, demonstrated a significant inverse correlation between molecular biology citations, and those for operative techniques, with the incidence of the latter dropping to 6% of all cited papers.

Discussion
This is the first study of its kind to analyse the most influential articles in hypospadiology, and their characteristics assessed for authorship, content and impact factor. Furthermore it allows data analysis by topic and between time points. This not only declutters the wealth of accumulated data on this subject, but also provides an important adjunct to education and training. As with this study, other citation analytical studies in urology found a significant preponderance in cited studies originating from the USA. The prevalence of the US in cited papers doesn't reflect any inherent bias, just a greater number of publications.

Conclusion
Although there are inherent potential elements of bias in citation analysis, this study demonstrates that citation analysis in a complicated topic can provide a high-throughput, uncomplicated method of quickly deciphering important contributions of authors and institutions to the field of hypospadias research.

http://dx.doi.org/10.1016/j.jpurol.2014.11.022

Introduction

As the field of paediatric urology has formed and matured, and with new surgical techniques providing outcomes that are believed to be better than those of the past, the literature related to hypospadias has grown substantially. Furthermore, the investigations of molecular mechanisms and possible environmental influences, although extensive, have not yet yielded any simple explanations.

Other issues have also been scrutinized, without consensus at this point in time, such as: self-esteem, the development of gender identity, body image from both the genital deformity and following reconstruction, reduced quality of life, depression and anxiety [1—4].

Despite the abundance of published literature, there remains a significant level of discordance in many aspects of hypospadiology. There is still no consensus on the aetiology of this condition, including molecular mechanisms and the associated environmental factors. Thirty-seven studies have been published within the last five years, of which 25 have focused on potential environmental factors including parental chemical exposure, parental characteristics, nutrition and hormones. The remaining papers have studied the hormone-dependent phase of molecular mechanism, namely: androgen and oestrogen-related genes. Of these, there have been no proven direct associations [5]. There are also data refuting an increased incidence in hypospadias, as part of a larger debate on the potential effects of so-called 'endocrine disruptors' such as phthalates and bisphenol-A, on male reproductive health [6].

There has been a volume of literature on the evolution of surgical techniques in hypospadias repair. The current goals of hypospadias repair include a functional penis that is adequate for sexual intercourse. Urethral reconstruction should offer the ability to stand to urinate and combine a satisfactory cosmetic result. Whether single-stage or two-stage procedures, or even with distal, less-complex urethral reconstructions, there is no consensus as to the ideal method of repair, some of which is attributed to non-uniform standards of reporting outcomes [7].

Given the prevalence of literature on hypospadiology, the issue of redundancy becomes apparent when results are duplicated in different publications by the same author/group, as well as indirect peer opinion as to the importance of a particular publication by way of citation frequency. The establishment of a citation rank list has been often used in medicine to identify peer-reviewed publications that have had the greatest intellectual influence [8]. When a peer-reviewed article references another publication, a citation is received. Citation analysis involves ranking and evaluating an article/journal based on the number of citations it receives. In addition to determining the most frequently cited articles, this analysis is also used to rank journals in terms of impact. Although the significance of citation analysis remains controversial, proponents argue that this method provides an objective method with which to determine the significance of an article or journal [9—11]. In the present study, this bibliographic analysis was undertaken to see where things stand today in the field that Sheldon and Duckett (1987) described as 'hypospadiology' [12]. It is the first study of its kind to analyse the most influential articles in the field of hypospadiology, and to assess their characteristics for authorship, content and impact factors.

Methods

To identify the most frequently cited articles published in hypospadiology, a cited reference search was carried out for indexed citations within the Web of Science™ Core Collection (Accessed 3rd February 2014) (Institutional Access; University of Dublin—Trinity College). The top 150 indexed, cited articles out of a total of 2457 citations containing the topic 'hypospadias' in the following subject categories were selected for analysis: urology/nephrology, urethra/development, paediatrics, surgery, genetics/hereditary and endocrinology/metabolism. Using a previously published protocol [10], the 'cited reference search' (a component of the Science Citation Index) command was used for each of these journals to identify the most frequently cited articles. References were evaluated for pertinence to the natural history, diagnosis, and/or management of hypospadias by two independent reviewers. The impact factors of the journals were cross-referenced with the 2012 edition of Journal Citation Reports (JCR): Science Edition (1945—2013). The impact factor of a journal is calculated based on the mean number of citations per year given to those papers in a journal that were published during the two preceding years.

In total, 48 journals were identified in the top 150 cited articles (10 urological/subspeciality and 38 medical/research). This database included publications from the 68-year period of 1945—2013. Each of the 48 journals was searched and every article with more than 40 citations was included in a comprehensive, ranked list.

The top 150 cited articles were compiled in the final list (Table 1). Complete inter-author agreement of the relevance led to the inclusion of a publication. Articles that were felt to be irrelevant to hypospadias or its management were excluded. The articles were accessed and reviewed online using MEDLINE. When relevant articles were unavailable online, they were received in print format via the University of Dublin, Trinity College library. The articles were reviewed and the data were tabulated according to the number of citations, country and institute of origin, journal, impact factor, authorship and topic.

Results

The mean number of citations for the top 150 articles was 76 (range 40—424), with articles on the list published between 1946 (Cecil et al., Journal of Urology) [13] and 2010 (Toppari et al., Birth Defects Res A) [14]. The top 150 articles were published from 21 countries, with 62% originating from the USA. The top three research institutions included the University of Texas (Southwestern) Medical Center, The University of California — San Francisco Medical Center, and the Children's Hospital — Philadelphia. In total, these represented 33 publications within the top 150 citations (22%) (Table 2). The most commonly utilised journal was the Journal of Urology, which published 58 of the top 150 cited hypospadias papers (39%). The most cited author

Table 1 The top 150 cited articles in hypospadiology (1945–2013).

Author	Title and citation	Year	Number of citations
Walsh PC et al.	Familial incomplete male pseudohermaphroditism, type-2 - decreased dihydrotestosterone formation in pseudovaginal perineoscrotal Hypospadias. NEJM **291**(18):944–9.	1974	424
Paulozzi IJ et al.	Hypospadias trends in two US surveillance systems. Pediatrics **100**(5):831–4.	1997	300
Snodgrass WT	Tubularized, incised plate urethroplasty for distal hypospadias. J Urol **151**(2):464–5.	1994	272
Paulozzi IJ	International trends in rates of hypospadias and cryptorchidism. Environ Health Perspect **107**(4):297–302.	1999	265
Duckett JW	MAGPI (meatoplasty and granuloplasty) - a procedure for sub-coronal hypospadias. Urol Clinics North Am **8**(3):513–9.	1981	178
Duckett JW	Transverse preputial island flap technique for repair of severe hypospadias. Urol Clinics North Am **7**(2):423–30.	1980	174
Sweet RA et al.	Study of incidence of hypospadias in Rochester, Minnesota, 1940–1970, and a case-control comparison of possible etiologic factors. Mayo Clin Proc **49**(1): 52–8.	1974	172
Welsh M et al.	Identification in rats of a programming window for reproductive tract masculinization, disruption of which leads to hypospadias and cryptorchidism. J Clin Invest **118**(4):1479–90.	2008	161
North K et al.	A maternal vegetarian diet in pregnancy is associated with hypospadias. BJU Int **85**(1):107–13.	2000	152
Toppari J et al.	Trends in the incidence of cryptorchidism and hypospadias and methodological limitations of registry-based data. Human Reprod Update **7**(3):282–6.	2001	140
Devine CJ et al.	A one stage hypospadias repair. J Urol **85**:166–72.	1961	140
Duckett JW	The island flap technique for hypospadias repair. Urol Clinics North Am **8**(3):503–11.	1981	139
Weidner IS et al.	Cryptorchidism and hypospadias in sons of gardeners and farmers. Envrion Health Perspect **106**(12):793–6.	1998	137
Baskin LS et al.	Hypospadias and endocrine disruption: is there a connection? Environ Health Perspect **109**(11):1175–83.	2001	132
Weidner IS et al.	Risk factors for cryptorchidism and hypospadias. J Urol **161**(5):1606–9.	1999	130
Akre O et al.	Risk factor patterns for cryptorchidism and hypospadias. Epidemiol **10**(4): 364–9.	1999	123
Devine CJ et al.	Hypospadias repair. J Urol **118**(1 Pt 2):188–93.	1977	123
Pierik FH et al.	Maternal and paternal risk factors for cryptorchidism and hypospadias: a case-control study in newborn boys. Environ Health Perspect **112**(15):1570–6.	2004	122
Snodgrass WT et al.	Tubularized incised plate hypospadias repair: results of a multicenter experience. J Urol **156**(2 Pt 2):839–41.	1996	122
Baskin LS et al.	Changing concepts of hypospadias curvature lead to more onlay island flap procedures. J Urol **151**(1):191–6.	1994	117
Baskin LS et al.	Anatomical studies of hypospadias. J Urol **160**(3 Pt 2):1108–15.	1998	114
Browne D	An operation for hypospadias. Proc R Soc Med **42**(7): 466–8.	1949	113
Klip H et al.	Hypospadias in sons of women exposed to diethylstilbestrol in utero: a cohort study. Lancet **359**(9312):1102–7.	2002	110
Snodgrass WT et al.	Tubularized incised plate hypospadias repair for proximal hypospadias. J Urol **159**(6):2129–31.	1998	110
Baskin LS	Hypospadias and urethral development. J Urol **163**(3):951–6.	2000	109
Elder JS et al.	Onlay island flap in the repair of mid and distal penile hypospadias without chordee. J Urol **138**(2):376–9.	1987	107
Boisen KA et al.	Hypospadias in a cohort of 1072 Danish newborn boys: prevalence and relationship to placental weight, anthropometrical measurements at birth, and reproductive hormone levels at three months of age. aJ Clin Endocrinol Metab **90**(7):4041–6.	2005	104
Longnecker MP et al.	Maternal serum level of 1,1-dichloro-2,2-bis(p-chlorophenyl)ethylene and risk of cryptorchidism, hypospadias, and polythelia among male offspring. Am J Epidemiol **155**(4):313–22.	2002	103
Atala A et al.	A novel inert collagen matrix for hypospadias repair. J Urol **162**(3 Pt 2):	1999	102

(*continued on next page*)

Table 1 (*continued*)

Author	Title and citation	Year	Number of citations
	1148—51.		
Dessanti A et al.	Autologous buccal mucosa graft for hypospadias repair - an initial report. J Urol **147**(4):1081—3.	1992	102
Kallen B et al.	A joint international study on the epidemiology of hypospadias. Acta Paediatr Scan Supp **324**:1—52.	1986	96
Baskin LS et al.	Dorsal tunica albuginea plication for hypospadias curvature. J Urol **151**(6): 1668—71.	1994	94
Aarskog D	Current concepts - maternal progestins as a possible cause of hypospadias. NEJM **300**(2):75—8.	1979	94
Bracka A	Hypospadias repair — the 2-stage alternative. Br J Urol **76** Suppl 3:31—41.	1995	93
Byars LT	A technique for consistently satisfactory repair of hypospadias. Surg Gynecol Obstet **100**(2):184—90.	1955	91
Borer JG et al.	Tubularized incised plate urethroplasty: expanded use in primary and repeat surgery for hypospadias. J Urol **165**(2):581—5.	2001	88
Silver RI et al.	In vitro fertilization is associated with an increased risk of hypospadias. J Urol **161**(6):1954—7.	1999	81
Baskin LS et al.	Hypospadias: anatomy, etiology, and technique. J Paediatr Surg **41**(3):463—72.	2006	80
Bracka A	A long-term view of hypospadias. Br J Plast Surg **42**(3):251—5.	1989	80
Hodgson NB	A one-stage hypospadias repair. J Urol **104**(2):281—3.	1970	79
King LR	Hypospadias - a one-stage repair without skin graft based on a new principle - chordee is sometimes produced by skin alone. J Urol **103**(5):660—2.	1970	75
Mureau MAM et al.	Psychosexual adjustment of children and adolescents after different types of hypospadias surgery - a norm-related study. J Urol **154**(5):1902—7.	1995	74
Walsh PC et al.	Plasma androgen response to hcg stimulation in prepubertal boys with hypospadias and cryptorchidism. J Clin Endocrinol Metab **42**(1):52—9.	1976	73
Asopa HS et al.	One stage correction of penile hypospadias using a foreskin tube. A preliminary report. Int Surg **55**(6):435—40.	1971	73
Hussain N et al.	Hypospadias and early gestation growth restriction in infants. Pediatrics **109**(3):473—8.	2002	72
Bowen P et al.	Hereditary male pseudohermaphroditism with hypogonadism hypospadias and gynecomastia (reifensteins syndrome). Ann Intern Med **62**:252—70.	1965	72
Mustarde JC	One-stage correction of distal hypospadias: and other people's fistulae. Br J Plastic Surg **18**(4):413—22.	1965	71
Fernandez MF et al.	Human exposure to endocrine-disrupting chemicals and prenatal risk factors for cryptorchidism and hypospadias: a nested case-control study. Environ Health Perspec **115** Suppl 1:8—14.	2007	70
Pierik FH et al.	A high hypospadias rate in the Netherlands. Human Reprod **17**(4):1112—5.	2002	70
Hollowell JG et al.	Preservation of the urethral plate in hypospadias repair - extended applications and further experience with the onlay island flap urethroplasty. J Urol **143**(1):98—100.	1990	70
Memmelaar J	Use of bladder mucosa in a one-stage repair of hypospadias. J Urol **58**(1): 68—73.	1947	69
Devine CJ et al.	Utricular configuration in hypospadias and intersex. J Urol **123**(3):407—11.	1980	66
Allera A et al.	Mutations of the androgen receptor coding sequence are infrequent in patients with isolated hypospadias. J Clin Endocrinol Metab **80**(9):2697—9.	1995	65
Barbagli G et al.	Failed hypospadias repair presenting in adults. Eur Urol **49**(5):887—94.	2006	64
Prosser DP et al.	Caudal tramadol for postoperative analgesia in paediatric hypospadias surgery. Br J Anaesth **79**(3):293—6.	1997	64
Romagnoli G et al.	Treatment of posterior hypospadias by the autologous graft of cultured urethral epithelium. NEJM **323**(8):527—30.	1990	64
Howard FS	Hypospadias with enlargement of the prostatic utricle. Surg Gynecol Obstet **86**(3):307—16.	1948	64
Felton LM	Should intravenous pyelography be a routine procedure for children with cryptorchism or hypospadias. J Urol **81**(2):335—38.	1959	63
Manson MC et al.	Molecular epidemiology of hypospadias: review of genetic and environmental risk factors. Birth Defects Res A Clin Mol Teratol **67**(10):825—36.	2003	61
Bracka A	A versatile 2-stage hypospadias repair. Br J Plast Surg **48**(6):345—52.	1995	61

Table 1 (*continued*)

Author	Title and citation	Year	Number of citations
Fredell L et al.	Heredity of hypospadias and the significance of low birth weight. J Urol **167**(3):1423—7.	2002	60
Baskin LS et al.	Urethral seam formation and hypospadias. Cell Tissue Res **305**(3):379—87.	2001	60
Mureau MAM et al.	Satisfaction with penile appearance after hypospadias surgery: patient and surgeon view. J Urol **155**(2):703—6.	1996	60
Retik AB et al.	Complications of hypospadias repair. Urol Clin North Am **15**(2):223—36.	1988	60
Calzolari E et al.	Etiologic factors in hypospadias. J Med Genet **23**:333—7.	1986	60
Culp OS	Struggles and triumphs with hypospadias and associated anomalies — review of 400 cases. J Urol **96**(3):339—51.	1966	60
Aschim EL et al.	Linkage between cryptorchidism, hypospadias, and ggn repeat length in the androgen receptor gene. J Clin Endocrinol Metab **89**(10):5105—9.	2004	59
Zaontz MR	The gap (glans approximation procedure) for glandular coronal hypospadias. J Urol **141**(2):359—61.	1989	59
Bauer SB et al.	Genetic-aspects of hypospadias. Urol Clinics North Am **8**(3):559—64.	1981	59
Shima H et al.	Developmental anomalies associated with hypospadias. J Urol **122**(5):619—21.	1979	59
Chen YC et al.	Genetic studies on hypospadias in males. J Med Genet **8**(2):153—9.	1971	59
Fukami M et al.	Cxorf6 is a causative gene for hypospadias. Nat Genet **38**(12):1369—71.	2006	58
Opitz JM	G-syndrome (hypertelorism with esophageal abnormality and hypospadias, or hypospadias-dysphagia, or opitz-frias or opitz-g syndrome) - perspective in 1987 and bibliography. Am J Med Genet **28**(2):275—85.	1987	58
Kallen B et al.	An epidemiological study of hypospadias in Sweden. Acta Paediatr Scand Supp. **293**:1—21.	1982	58
Secrest CL et al.	Repair of the complications of hypospadias surgery. J Urol **150**(5 Pt 1):1415—8.	1993	57
Berg R et al.	Social and sexual adjustment of men operated for hypospadias during childhood - a controlled-study. J Urol **125**(3):313—7.	1981	57
Czeizel A et al.	Etiological studies of hypospadias in Hungary. Hum Hered **29**(3):166—71.	1979	57
Roberts CJ et al.	Observations on epidemiology of simple hypospadias. BMJ **1**(5856):768—70.	1973	57
Goldman AS et al.	Production of congenital adrenal cortical hyperplasia hypospadias and clitoral hypertrophy (adrenogenital syndrome) in rats by inactivation of 3beta-hydroxysteroid dehydrogenase. Proc Soc Exp Biol Med **121**(3):757—66.	1966	57
Retik AB et al.	Meatal based hypospadias repair with the use of a dorsal subcutaneous flap to prevent urethrocutaneous fistula. J Urol **152**(4):1229—31.	1994	56
Koff SA	Mobilization of the urethra in the surgical-treatment of hypospadias. J Urol **125**(3):394—7.	1981	56
Khuri FJ et al.	Urologic anomalies associated with hypospadias. Urol Clinics North Am **8**(3): 565—71.	1981	56
Browne D	Hypospadias. Postgrad Med J **25**(286): 367—72.	1949	56
Silver RI et al.	5 alpha-reductase type 2 mutations are present in some boys with isolated hypospadias. J Urol **162**(3 Pt 2):1142—5.	1999	55
Sutherland RW et al.	Androgen receptor gene mutations are rarely associated with isolated penile hypospadias J. Urol. **156**(2 Pt 2):828—31	1996	55
Hiort O et al.	Molecular characterization of the androgen receptor gene in boys with hypospadias. Eur J Pediatr **153**(5):317—21.	1994	55
Duckett JW et al.	Meatal advancement and glanuloplasty hypospadias repair after 1000 cases - avoidance of meatal stenosis and regression. J Urol **147**(3):665—9.	1992	55
Rich MA et al.	Hinging the urethral plate in hypospadias meatoplasty. J Urol **142**(6):1551—3.	1989	55
Devine CJ et al.	Chordee without hypospadias. J Urol **110**(2):264—71.	1973	55
Snodgrass WT et al.	Tubularized incised-plate urethroplasty for proximal hypospadias. BJU Int **89**(1):90—3.	2002	54
Snodgrass WT et al.	Tubularized incised-plate urethroplasty for hypospadias reoperation. B JU Int **89**(1):98—100.	2002	54
Christiansen S et al.	Combined exposure to anti-androgens causes markedly increased frequencies of hypospadias in the rat. Int J Androl **31**(2):241—8.	2008	53
Baskin LS et al.	Buccal mucosa grafts in hypospadias surgery. Br J Urol **76** Suppl 3:23—30.	1995	53
Snow BW	Use of tunica vaginalis to prevent fistulas in hypospadias surgery. J Urol **136**(4):861—3.	1986	53

(*continued on next page*)

Table 1 (*continued*)

Author	Title and citation	Year	Number of citations
Migeon CJ et al.	Ambiguous genitalia with perineoscrotal hypospadias in 46, xy individuals: long-term medical, surgical, and psychosexual outcomes. Pediatrics **110**(3):e31.	2002	52
Aho M et al.	Is the incidence of hypospadias increasing? Analysis of Finnish hospital discharge data 1970–1994. Environ Health Perspect **108**(5):463–5.	2000	52
Snodgrass WT	Does tubularized incised plate hypospadias repair create neourethral strictures? J. Urol. **162**(3 Pt 2):1159–61	1999	52
Dolk H	Rise in prevalence of hypospadias. Lancet **351**(9105):770.	1998	52
Steckler RE et al.	Stent-free Thiersch-Duplay hypospadias repair with the Snodgrass modification. J Urol **158**(3 Pt 2):1178–80.	1997	52
Albers N et al.	Etiologic classification of severe hypospadias: implications for prognosis and management J. Paediatr. **131**(3):386–92.	1997	52
Mureau MAM et al.	Psychosexual adjustment of men who underwent hypospadias repair - a norm-related study. J Urol **154**(4):1351–5.	1995	52
Rajfer J et al.	Incidence of intersexuality in patients with hypospadias and cryptorchidism. J Urol **116**(6):769–70.	1976	52
Brouwers MM et al.	Hypospadias: a transgenerational effect of diethylstilbestrol? Human Reprod **21**(3):666–9.	2006	51
Allen TD et al.	Surgical treatment of coronal hypospadias and related problems. J Urol **100**(4):504–8.	1968	51
Snodgrass WT	Tubularized incised plate hypospadias repair: indications, technique, and complications. Urology **54**(1):6–11.	1999	50
Retik AB et al.	Management of severe hypospadias with a 2-stage repair. J Urol **152**(2 Pt 2):749–51.	1994	50
Allen TD et al.	Endocrine studies in patients with advanced hypospadias. J Urol **131**(2):310–4.	1984	50
Erol A et al.	Anatomical studies of the urethral plate: why preservation of the urethral plate is important in hypospadias repair. BJU Int **85**(6):728–34.	2000	49
Perovic S et al.	Onlay island flap urethroplasty for severe hypospadias - a variant of the technique. J Urol **151**(3):711–4.	1994	49
Carmichael SL et al.	Hypospadias in California - trends and descriptive epidemiology. Epidemiol **14**(6):701–6.	2003	48
Fisch H et al.	Maternal age as a risk factor for hypospadias. J Urol. **165**(3):934–6	2001	48
Firlit CF	The mucosal collar in hypospadias surgery J Urol **137**(1):80–2.	1987	48
Ormond G et al.	Endocrine disruptors in the workplace, hair spray, folate supplementation, and risk of hypospadias: case-control study. Environ Health Perspect **117**(2):303–7.	2009	47
Carmichael SL et al.	Maternal progestin intake and risk of hypospadias. Arch Pediatr Adolesc Med **159**(10):957–62.	2005	47
Hensle TW et al.	Buccal mucosa grafts for hypospadias surgery: long-term results. J Urol **168**(4 Pt 2):1734–6.	2002	47
Mouriquand PDE et al.	Hypospadias repair - current principles and procedures. Br J Urol **76** Suppl 3:9–22.	1995	47
Manzoni G et al.	Hypospadias surgery: when, what and by whom? BJU Int **94**(8):1188–95.	2004	46
Kim KS et al.	Induction of hypospadias in a murine model by maternal exposure to synthetic estrogens. Environ Res **94**(3):267–75.	2004	46
Dolk H et al.	Toward the effective surveillance of hypospadias. Environ Health Perspect **112**(3):398–402.	2004	46
Cheng EY et al.	Snodgrass hypospadias repair with vascularized dartos flap: the perfect repair for virgin cases of hypospadias? J Urol **168**(4 Pt 2):1723–6.	2002	46
Mollard P et al.	Hypospadias - the release of chordee without dividing the urethral plate and onlay island flap (92 cases). J Urol **152**(4):1238–40.	1994	46
Belman AB	De-epithelialized skin flap coverage in hypospadias repair. J Urol **140**(5 Pt 2):1273–6.	1988	46
Schultz JR et al.	Timing of elective hypospadias repair in children. Pediatrics **71**(3):342–51.	1983	46
Hsieh MH et al.	Associations among hypospadias, cryptorchidism, anogenital distance, and endocrine disruption. Curr Urol Rep **9**(2):137–42.	2008	45
Wang YP et al.	Mutation analysis of five candidate genes in Chinese patients with hypospadias. Eur J Human Genet **12**(9):706–12.	2004	45

Table 1 (*continued*)

Author	Title and citation	Year	Number of citations
Van Tongeren M et al.	A job-exposure matrix for potential endocrine-disrupting chemicals developed for a study into the association between maternal occupational exposure and hypospadias. Ann Occup Hyg **46**(5):465–77.	2002	45
Opitz JM et al.	Pseudovaginal perineoscrotal hypospadias. Clin Genet **3**(1):1–26.	1972	45
Muldal S et al.	Deletion of y chromosome in a family with muscular dystrophy and hypospadias. BMJ **1**(5274):291–4.	1962	45
Wang M-H et al.	Endocrine disruptors, genital development, and hypospadias. J Androl **29**(5):499–505.	2008	44
Brouwers MM et al.	Risk factors for hypospadias. Eur J Paediatr **166**(7):671–8.	2007	44
Snodgrass WT et al.	Initial experience with staged buccal graft (bracka) hypospadias reoperations. J Urol **172**(4 Pt 2):1720–4.	2004	44
Snodgrass WT et al.	Histology of the urethral plate: implications for hypospadias repair. J Urol **164**(3 Pt 2):988–9.	2000	44
Stoll C et al.	Genetic and environmental-factors in hypospadias. J Med Genet **27**(9):559–63.	1990	44
Aarskog D	Intersex conditions masquerading as simple hypospadias. Birth Defects Orig Artic Ser **7**(6):122–30.	1971	44
Broadbent T et al.	Hypospadias - one-stage repair. Br J Plast Surg **18**(4):406–12.	1965	44
Rey RA et al.	Low risk of impaired testicular sertoli and leydig cell functions in boys with isolated hypospadias. J Clin Endocrinol Metab **90**(11):6035–40.	2005	43
Retik AB et al.	Primary and reoperative hypospadias repair with the Snodgrass technique. World J Urol **16**(3):186–91.	1998	43
Svensson J et al.	Androgen receptor levels in preputial skin from boys with hypospadias. J Endocrinol Metab **49**(3):340–5.	1979	43
Cecil AB	Repair of hypospadias and urethral fistula. J Urol **56**(2):237–42.	1946	43
Snodgrass WT et al.	Current technique of tubularized incised plate hypospadias repair. Urology **60**(1):157–62.	2002	42
Hensle TW et al.	Hypospadias repair in adults: adventures and misadventures. J Urol **165**(1): 77–9.	2001	42
Hakim S et al.	Outcome analysis of the modified Mathieu hypospadias repair: comparison of stented and unstented repairs. J Urol **156**(2 Pt 2):836–8.	1996	42
Toppari J et al.	Cryptorchidism and hypospadias as a sign of testicular dysgenesis syndrome (tds): environmental connection. Birth Defects Res A Clin Mol Teratol **88**(10):910–9.	2010	41
Shanberg AM et al.	Re-operative hypospadias repair using the Snodgrass incised plate urethroplasty. BJU Int **87**(6):544–7.	2001	41
Fredell L et al.	Hypospadias is related to birth weight in discordant monozygotic twins. J Urol **160**(6 Pt 1):2197–9.	1998	41
Rabinowitz R	Outpatient catheterless modified Mathieu hypospadias repair. J Urol **138**(4 Pt 2):1074–6.	1987	41
Hayes HM et al.	Hospital incidence of hypospadias in dogs in north America. Vet Rec **118**(22):605–7.	1986	41
Backus LH et al.	Hypospadias-then and now. Plast Reconstr Surg Transplant Bull **25**:146–60.	1960	41
Gatti JM et al.	Increased incidence of hypospadias in small-for-gestational age infants in a neonatal intensive-care unit. BJU Int **87**(6):548–50.	2001	40
Creevy CD	The correction of hypospadias: a review. Urol Surv **8**(1):2–47.	1958	40

was W.T. Snodgrass, with 10 citations within the top 150 papers, followed by L.S. Baskin, with eight citations. There were a total of 23 authors with two or more citations within the top 150 articles (Table 3).

There has been a steady increase in the number of publications in the field of hypospadiology over the last eight decades, which is mirrored in citation analysis. There were five citations from the period 1940–1950 (3%) compared with 49 citations from 2000 to 2010 (33%) (Fig. 1). The majority of citations (67 papers) dealt with the topic of surgical technique and surgical management of hypospadias (45%). This was followed by the impact of environmental influence on the development of hypospadias (21%), molecular biology of hypospadias (16%), epidemiology and trends associated with hypospadias development (15%), and psychosexual aspects (3%).

Table 2 Institution and country of origin of the 150 most frequently cited hypospadias publications (1945—2013) (Abbreviations: MC — Medical Centre; NY — New York; UK — United Kingdom).

Institution	Hypospadias citations	Country	Hypospadias citations
University of Texas MC (Southwestern)	**13**	USA	**93**
University of California, San Francisco MC	**11**	UK	**14**
Children's Hospital, Philadelphia	**9**	Sweden	**8**
Boston Children's Hospital	**7**	Netherlands	**8**
Johns Hopkins Hospital, Maryland	**7**	Italy	**5**
Karolinska Institute, Stockholm	**5**	Denmark	**4**
Sentara Norfolk General Hospital, Virginia	**5**	Norway	**3**
Columbia University MC, NY	**3**	Finland	**2**
Erasmus University MC, Rotterdam	**3**	France	**2**
Great Ormond St. Hospital, London	**3**	Germany	**2**
Wordsley Hospital, Stourbridge, UK	**3**	Japan	**2**

Discussion

Hypospadias research involves multiple specialities that encompass ideas and research from reconstructive surgery, epidemiology, molecular biology and psychological medicine. Major contributions to this field have helped shape modern treatment protocols and improved outcomes for people with hypospadias. Recognition of this work is important in helping physicians and scientists understand and design future research.

The top 150 cited articles in hypospadiology, from 48 recognised peer-reviewed journals, were identified. Citation analysis was chosen as the determinant of selection. Although there is no direct correlation between citation frequency and study quality, it offers an insight into the degree of peer analysis, the readership of the manuscript and a measure of recognition. It also highlights an acceptance of their importance in the field. An awareness of these articles, mainly in high-impact journals, can also educate trainees regarding research methodology and ethical considerations.

Many of these subspecialty articles were published in high-impact-factor journals, with a median impact factor of 4.18 across the 17 most highly cited journals (range 1.23—51.66). There were only eight articles in the top 150 that were not associated with original research (5%); therefore, the volume of generated data is encouraging. The most productive decade of hypospadias research in the top 150 was 2000—2010, with 49 citations. There has been a linear increase in the number of papers published since the 1940s, which suggests the most influential contributions in hypospadias research are relatively recent. This could in turn be attributed to a more recent rapid evolution in the awareness, treatment, and quality of research for this disorder. It is no surprise that the majority of top cited papers in hypospadiology originate from US institutions (64%). This follows patterns from urology, anaesthesia, orthopaedics and general surgery [10,11,15—17]. The size and

Table 3 Journal, impact factor and top cited authors of the 150 most frequently cited hypospadias publications (1945—2013).

Journal	Total citations	Impact factor (2012)	First author	Total citations
Journal of Urology	**58**	**3.696**	Snodgrass WT	**10**
BJU International (British Journal of Urology)	**10**	**3.046**	Baskin LS	**8**
Environmental Health Perspectives	**8**	**7.260**	Devine CJ	**4**
Journal of Clinical Endocrinology & Metabolism	**6**	**6.430**	Duckett JW	**4**
Urologic Clinics of North America	**6**	**1.385**	Retik AB	**4**
Pediatrics	**4**	**5.119**	Bracka A	**3**
British Journal of Plastic Surgery	**3**	**1.290**	Mureau MAM	**3**
Journal of Medical Genetics	**3**	**5.703**	Aarskog D	**2**
New England Journal of Medicine	**3**	**51.658**	Allen TD	**2**
Acta Paediatrica (Scandinavica)	**2**	**1.974**	Brouwers MM	**2**
Birth Defects Research Part A - Clinical and Molecular Teratology	**2**	**3.146**	Browne D	**2**
British Medical Journal	**2**	**17.215**	Carmichael SL	**2**
Epidemiology	**2**	**5.738**	Dolk H	**2**
European Journal of Pediatrics	**2**	**1.907**	Fredell L	**2**
Human Reproduction	**2**	**4.670**	Hensle TW	**2**
The Lancet	**2**	**39.060**	Kallen B	**2**
Surgery, Gynecology & Obstetrics	**2**	**1.225**	Opitz JM	**2**
Urology	**2**	**2.420**	Paulozzi IJ	**2**

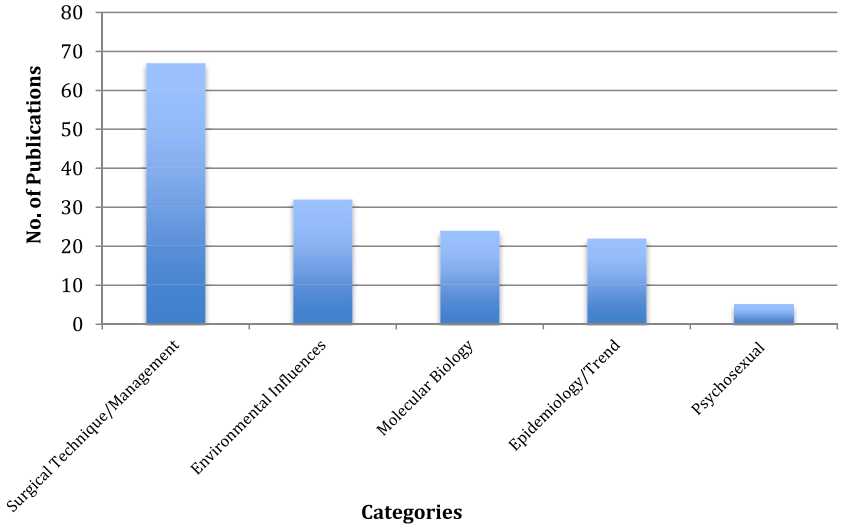

Figure 1 Linear analysis of the total number of citations in hypospadiology grouped by decade (1945—2013).

funding opportunities for US-led scientific and clinical research is greater than elsewhere, and promotes high publication output.

Analysing the entire list of 150 top-cited articles in hypospadiology lies outside the scope of the present article, but a number of observations can be made from this list. Eight of the top twenty articles deal with surgical technique, with the most commonly cited paper being the description of the tubularised, incised plate urethroplasty by Snodgrass et al., in 1994 (272 citations) [18]. The top cited molecular biology paper is by Walsh et al., in 1974 (424 citations), it describes familial incomplete male pseudohermaphroditism and the development of pseudovaginal, perineoscrotal hypospadias [19]. Clearly, there have been a significant number of important, even landmark, papers since the publication of the Walsh paper, and one of the drawbacks that this analysis doesn't take into account is that seminal papers can serve to discover new breakthroughs and talents more quickly than classical citation indices. An argument could also be made that citation analysis also fails to outline the practical importance of key peer-reviewed publications. Seven of the top twenty articles originate from Europe (35%). The top ranked of these by citation is by Welsh et al., in 2008 (UK), it identifies a programming window for reproductive tract masculinization in rats [20]. Although the vast majority of papers originate from the US, a significant proportion of influential contributions also emanate from Europe.

There have been a number of studies exploring the value of citation analyses in the field of urology. Both Nason et al. [10] and Hennessey et al. [16] analysed the top 100 articles in the field of urology. Observational studies and randomised, controlled trials in oncology published in high-impact journals constituted the most common type of highly cited publications. In the Hennessey study, there was a mean citation of 629, with 54% published prior to the 1990s, followed by a mean citation of 892 by Nason et al. with an overall 19% change in the top 100 articles over five years. As with the present study, both found a significant preponderance in cited studies originating from the USA. Both studies also illustrated a weighting toward oncology and transplantation as the most highly cited topics. Li et al. performed a comparative study of scientific publications originating from China, the US and Japan, and found a significant increase in the number of cited articles over a ten-year period, and that articles originating from the USA had the highest impact factors, citations and randomised, controlled trials ($P < 0.001$) [21]. This highlights the variable nature of citation analysis, with higher proportions cited in more recent years. The prevalence of the US in cited papers doesn't reflect any inherent bias, just a greater number of publications; this was demonstrated in the present study as well.

There are a number of differences in the patterns of publications over each decade. A total of 39% papers in the top 150 cited articles were published prior to 1990. Three out of the five papers cited in the 1940s dealt with operative techniques to repair hypospadias, followed by a further paper in the 1950s. The 1947 paper by Memmelaar et al. that seemed to have been forgotten was resurrected in the 1990s (and also misnamed 'bladder mucosal' graft). The 1948 paper by Howard on utricles seemed to add comparatively little to hypospadiology, with respect to larger papers from later decades; however, it remained highly cited. Brown et al. was cited twice in 1949. The 1960s featured nine articles, two of which were reviews, and for the first time, three cited articles on molecular biology. There were also two original articles on operative technique by Broadbent, and by Devine. There is then an accelerated increase in the number of cited articles from the 1970s to the present. One of the most striking features of citation analysis is the difference in the frequency of descriptions of original techniques for repair over the decades. As the prevalence in molecular biology, psychology and environmental factors of hypospadiology increases, there has been a steady decline in the comparative description of new surgical techniques. There have been 28 papers in the top 150 cited (19%) that deal with the description of new techniques. In the 1940—1950s, this constituted 60% of cited publications. Over the next two decades, new techniques were cited at a frequency of 33% compared with overall publications. The 1970s and 1980s saw a further

drop in cited techniques to 20% overall cited output; however, this period gave rise to a number of seminal papers including the MAGPI repair (Duckett 1980) and the preputial onlay island-flap (Duckett 1981). Snodgrass, Bracka and Retik feature heavily in the 1990s, with a number of cited publications; however, overall technique description falls to 23% (although remains highest with 10 papers). The period 2000—2010 demonstrates a significant inverse correlation between molecular biology citations and those for operative techniques, with the incidence of the latter dropping to 6% of all cited papers.

There were limitations to the present study. One was that the Science Citation Index Expanded did not track citations for articles published prior to 1945 and, therefore, omissions of important citations prior to this date, although unlikely, are possible. Other potential criticisms of citation analysis, despite providing an objective and quantitative measure of the impact of an article, is that it fails to account for self-citation and language bias. Nor does it distinguish between positive or negative citations. A paper that is repeatedly cited because it was flawed or subsequently disproved could theoretically rank highly in a bibliometric analysis such as this [22—25]. It should also be noted that due to the inherent limitations in citation analysis, as discussed above, there would inevitably be an omission of some papers that many experts would consider essential to the field. These seminal papers would be included in a review or meta-analysis of this field; however, the identification of these papers is not the aim of a citation analysis.

A further bias is the 'obliteration by incorporation' effect described by Garfield et al. (1987) [26]. This refers to the existing phenomenon where older publications are no longer cited with the same frequency, as their findings become incorporated into the field's current body of knowledge. Due to this effect, this analysis ranked the articles based on the total number of citations received as opposed to the number of citations received in the current year. If the number of citations in the current year were used as the ranking criteria, this would produce an unfair relative increase in citations for more recent articles, as opposed to fewer citations for many 'classic articles' that have an earlier publication date. There is also a time lag for influential papers to accumulate enough citations to potentially warrant inclusion. For this reason, it is felt that this process should be repeated to update the top 150 most cited articles in hypospadiology on an ongoing basis, and to determine any changes, if applicable, to this list [21].

Conclusion

It is believed that this is the first study of its kind to examine citation analyses and the trends associated with hypospadias publishing. There is an ever-increasing amount of research associated with the different facets of hypospadiology, and it can be difficult to initially identify the significance of particular publications in a field with much uncertainty.

Citation analysis provides an objective and quantitative measure of the impact that an article has on its respective field, and can help to demystify some of the uncertainty associated with controversial topics such as hypospadiology over the last eight decades. This technique should allow researchers to optimise the time spent on literature analysis and assist with future research/investigative efforts.

Conflict of interest/funding

None declared.

Disclosure

None of the authors has direct or indirect commercial financial incentives associated with publishing the present article. They do not have any conflict of interest, and have signed the Disclosure Form.

References

[1] Schönbucher VB, Landolt MA, Gobet R, Weber DM. Psychosexual development of children and adolescents with hypospadias. J Sex Med 2008;5:1365—73.
[2] Eray N, Dogangun B, Kayaalp L, Emir H, Soylet Y, Danismend N, et al. Emotional effects of hypospadias surgery on Turkish boys. J Pediatr Urol 2005;1:75—80.
[3] Berg R, Svensson J, Aström G. Social and sexual adjustment of men operated for hypospadias during childhood: a controlled study. J Urol 1981;125:313—7.
[4] Yang Q, Qu WY, Yang L, Wang K, Tu HY, Wang J. Literature on the aetiology of hypospadias in the last 5 years: molecular mechanism and environmental factors. Andrologia 2014;46(6):583—91.
[5] Fisch H, Hyun G, Hensle TW. Rising hypospadias rates: disproving a myth. J Pediatr Urol 2010;6:37—9.
[6] Hayashi Y, Kojima Y. Current concepts in hypospadias surgery. Int J Urol 2008;15:651—64.
[7] Murray MR, Wang T, Schroeder GD, Hsu WK. The 100 most cited spine articles. Eur Spine J 2012;21:2059—69.
[8] Taubes G. Measure for measure in science. Science 1993;260:884—6.
[9] Brody S. Impact factor as the best operational measure of medical journals. Lancet 1995;346:1300—1.
[10] Nason GJ, Tareen F, Mortell A. The top 100 cited articles in urology: an update. Can Urol Assoc J 2013;7:E16—24.
[11] Lefaivre KA, Shadgan B, O'Brien PJ. 100 most cited articles in orthopaedic surgery. Clin Orthop Relat Res 2011;469:1487—97.
[12] Sheldon C, Duckett J. Hypospadias. Paed Clin N. Am 1087; 34:1259—1272.
[13] Cecil AB. Repair of hypospadias and urethral fistula. J Urol 1946;56:237—42.
[14] Toppari J, Virtanen HE, Main KM, Skakkebaek NE. Cryptorchidism and hypospadias as a sign of testicular dysgenesis syndrome (TDS): environmental connection. Birth Defects Res A Clin Mol Teratol 2010;88:910—9.
[15] Uthman OA, Okwundu CI, Wiysonge CS, Young T, Clarke A. Citation classics in systematic reviews and meta-analyses: who wrote the top 100 most cited articles? PLoS One 2013;8:e78517.
[16] Hennessey K, Afshar K, Macneily AE. The top 100 cited articles in urology. Can Urol Assoc J 2009;3:293—302.
[17] Paladugu R, Schein M, Gardezi S, Wise L. One hundred citation classics in general surgical journals. World J Surg 2002;26:1099—105.
[18] Snodgrass W. Tubularized, incised plate urethroplasty for distal hypospadias. J Urol 1994;151:464—5.

[19] Walsh PC, Madden JD, Harrod MJ, Goldstein JL, MacDonald PC, Wilson JD. Familial incomplete male pseudohermaphroditism, type 2. Decreased dihydrotestosterone formation in pseudovaginal perineoscrotal hypospadias. N Engl J Med 1974;291:944—9.

[20] Welsh M, Saunders PT, Fisken M, Scott HM, Hutchinson GR, Smith LB, et al. Identification in rats of a programming window for reproductive tract masculinization, disruption of which leads to hypospadias and cryptorchidism. J Clin Invest 2008;118:1479—90.

[21] Li J, Gao XH, Bian Q, Guo ZY, Mei XB, Yu G, et al. Comparative study of scientific publications in urology and nephrology journals originating from USA, China and Japan (2001-2010). PLoS One 2012;7:e42200.

[22] Weale AR, Bailey M, Lear PA. The level of non-citation of articles within a journal as a measure of quality: a comparison to the impact factor. BMC Med Res Methodol 2004;4:14.

[23] Bakkalbasi N, Bauer K, Glover J, Wang L. Three options for citation tracking: Google Scholar, Scopus and Web of Science. Biomed Digit Libr 2006;3:7.

[24] Filion KB, Pless IB. Factors related to the frequency of citation of epidemiologic publications. Epidemiol Perspect Innov 2008;5:3.

[25] Callaham M, Wears RL, Weber E. Journal prestige, publication bias, and other characteristics associated with citation of published studies in peer-reviewed journals. JAMA 2002;287:2847—50.

[26] Garfield E. 100 citation classics from the Journal of the American Medical Association. JAMA 1987;257:52—9.