Journal of Pediatric Urology (2016) 12, 152.e1–152.e7

Worldwide prevalence of hypospadias

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Keywords
Hypospadias; Incidence; Prevalence; Epidemiology

Received 1 May 2015
Accepted 1 December 2015
Available online 31 December 2015

Summary

Introduction
Hypospadias is a common congenital malformation. Surgical repair and management of the long-term consequences require a substantial amount of socioeconomic resources. It is generally accepted that genetic and environmental factors play a major role in the etiology of hypospadias. There have been contradictory reports on rising hypospadias rates, and regional and ethnical differences. The exact prevalence of hypospadias is of major interest for healthcare providers, clinical medicine, and research.

Objective
To review the literature regarding the worldwide prevalence of hypospadias.

Study design
Pubmed, EMBASE and Google were systematically screened for: hypospadias, congenital malformation, anomaly, incidence, prevalence, and epidemiology. Exclusion criteria were surgical and risk-factor studies. To give an additional comprehensive overview, prevalence data were harvested from the Annual Report of the International Clearinghouse Centre for Birth Defects Surveillance and Research. Prevalence was reported as per 10,000 live births.

Results
Data were available from 1910 to 2013. The median study period was 9 years (range: 1–36 years). Approximately 90,255,200 births have been screened in all studies. The mean prevalence were: Europe 19.9 (range: 1–464), North America 34.2 (6–129.8), South America 5.2 (2.8–110), Asia 0.6–69, Africa 5.9 (1.9–110), and Australia 17.1–34.8. There were major geographical, regional, and ethnical differences, with an extreme heterogeneity of published studies. Numerous studies showed an increasing prevalence; on the other hand, there were a lot of contradictory data on the prevalence of hypospadias. The summary table shows contradictory data from the five largest international studies available.

Discussion
There was huge literature available on the prevalence of hypospadias. Most data derived from Europe and North America. Many methodological factors influenced the calculation of an accurate prevalence, and even more of the true changes in prevalence over time (no generally accepted and standardized definition of hypospadias, different monitoring systems, unclear efficiency of notification and data ascertainment, etc.). There was wide variation of prevalence according to countries and ethnicity, and there were conflicting data on the recent trends of prevalence. Moreover, there weren't any epidemiologic data available from many parts of the world.

Conclusion
True prevalence of hypospadias and trends were difficult to estimate. For the future, to be able to assess the true prevalence of hypospadias and changes in prevalence collaboration of national and international prospective registers is recommended.

Study	Year	Country	Per 10,000 live births	Time period	Trend
Bergman et al.	2015	23 European regions	18.6	2001–2010	**Different trends**
Chul Kim et al.	2011	South Korea	1.4–3.3	2000–2004	**Increasing**
Elliott et al.	2011	USA	42	1985–2006	**No increase**
Paulozzi	1999	International registries	10–45	1964–1997	**Partial increase**
Paulozzi et al.	1997	USA	17–50	1968–1993 1970–1993	**Increasing**

http://dx.doi.org/10.1016/j.jpurol.2015.12.002

Introduction

Hypospadias is the most common congenital malformation of the penis, and it is considered by some to be a mild form of 46XY disorders of sex development. The urethral opening occurs on the ventral side of the penis; moreover, there is hypoplasia of the ventral tissues in varying degrees, penile curvature, and excessive prepuce. It is generally accepted that genetic and environmental factors play a major role in the etiology of hypospadias [1]. Surgical repair of hypospadias and management of the long-term consequences of being born with a genital condition require socioeconomic resources. Therefore, the exact prevalence of hypospadias is of major interest for the community. Recently, the epidemiology of hypospadias and whether the prevalence is rising or stable has become part of a major debate over the potential effects of environmental factors [2]. This article systematically reviewed the literature regarding the worldwide prevalence of hypospadias and changes of prevalence over time.

Methods

Pubmed, EMBASE, and Google (to March 2015) were screened for potentially eligible literature using the following keywords: ((hypospadias) AND epidemiology) OR ((hypospadias) and incidence) OR ((hypospadias) and prevalence) (Fig. 1). The Related Article feature of PubMed was used and the search was restricted to articles in English. For countries or regions with no articles on hypospadias the search was extended with the terms congenital malformation, and anomaly. Exclusion criteria were: surgical series or surgical outcome analysis, reviews, case reports, obviously recycled data, and data of dubious quality. Moreover, data were not included from articles focusing on the etiology of hypospadias, genetics, risk factors or epidemiologic casual research (environmental factors, maternal exposure, etc.), unless prevalence data were clearly extractable. The authors screened the full text for eligibility, when available. Data collected were: authors, date of publication, study country or region, size of study group/cohort, prevalence of hypospadias, mild/severe ratio of hypospadias, trend, and study design or database/register. To give an additional comprehensive overview, prevalence data were harvested from the Annual report of the International Clearinghouse Centre for Birth Defects Surveillance and Research (ICBDSR) [3].

Prevalence is the proportion of a study population found to have a condition, and is expressed as the number of cases per 1000, 10,000 or 100,000 people. There is no standardized prevalence and authors use different measurements like per 1000, per 10,000, per 10,000 live births, per 10,000 live births including miscarriage and still births, per 1000 male births, and so on. This makes numbers extraordinary difficult to compare; therefore, the present study stated prevalence per 10,000 live births. In some studies, prevalence had to be estimated/calculated by assuming a male:female ratio of 50:50. The mean prevalence per geographic region was calculated by total number of hypospadias/total number of births x 10,000.

Results

Fig. 2 shows the worldwide available studies on the prevalence of hypospadias, including the mean, minimum and

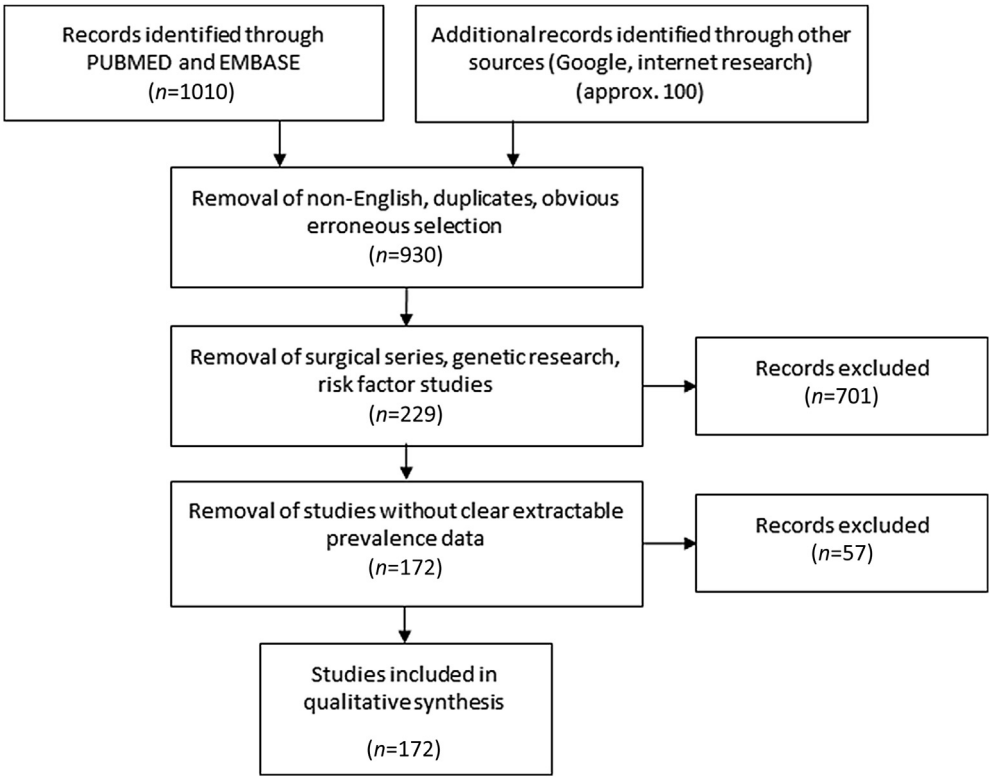

Figure 1 Systematic literature research.