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INCIDENCE AND SURGICAL REPAIR RATES OF HYPOSPADIAS IN NEW YORK STATE

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ABSTRACT

Objectives. To determine the trend of hypospadias incidence in New York State (NYS) by retrospectively evaluating both the incidence and surgical repair rates of hypospadias. Epidemiologic studies in the United States and Europe have suggested an observable trend in the incidence of hypospadias during the past 30 years. A comparison of the results from these studies, however, revealed inconsistent trends.

Methods. The incidence and surgical repair rates of hypospadias between 1983 and 1995 were extracted from the Congenital Malformations Registry and State Wide Planning Research Cooperative System of NYS. Pearson's correlation coefficient was used to determine the direction and magnitude of change.

Results. Neither the incidence nor the repair rates of hypospadias in NYS between 1983 and 1995 changed significantly during the course of these 12 years ($r = -0.225$, $P = 0.45$ and $r = -0.010$, $P = 0.92$, respectively).

Conclusions. The results demonstrated no statistically observable trend in either the incidence or surgical repair rates of hypospadias in NYS between 1983 and 1995. UROLOGY **57:** 151–153, 2001. © 2001, Elsevier Science Inc.

Hypospadias is a common congenital anomaly that occurs in 1 of every 300 live male births.[1] Epidemiologic studies in the United States and Europe have suggested an observable trend in the incidence of hypospadias during the past 30 years.[2–5] A comparison of the results from these studies, however, revealed inconsistent trends. Data from the Metropolitan Atlanta Congenital Defects Program (MACDP) showed a twofold increase in the hypospadias incidence rates. Data from the California Birth Defects Monitoring Program, however, revealed no significant changes. The Italian Multicentric Register of Congenital Malformations demonstrated a rise in incidence, but the congenital malformation registry in Spain showed a decline. The reason for this disparity in the incidence rate of hypospadias is unclear.

Researchers have suggested that this disparity may be due to different degree of exposure to endocrine-disrupting environmental pollutants (ie, industrial chemicals) and medications (ie, contraceptives).[6–8] Others have suggested that it may be due to different diagnostic criteria for the minor forms of hypospadias in which the meatal opening is within the glans penis.

We decided to determine the trend of the hypospadias incidence in New York State (NYS) by retrospectively evaluating both the incidence and surgical repair rates of hypospadias.

MATERIAL AND METHODS

The incidence of hypospadias between 1983 and 1995 was obtained from the Congenital Malformation Registry of NYS, which was established in 1982. The rate of surgical repair of hypospadias during the same period was obtained from the hospital discharge data set of NYS, the State Wide Planning Research Cooperative System.

Birth defects refer to "congenital anomalies" as identified by codes 740–759 of the Ninth Revision of the International Classification of Diseases. Hypospadias was identified by code 752.6. The incidence includes only live births. Most of the reported data for incidence includes children diagnosed after 2 weeks old and before 2 months old.

The State Wide Planning Research Cooperative System data of NYS identified each repair by the International Classification of Diseases procedure codes. Hypospadias repair was identified by code 58.45. Approximately 70% of the procedures were performed in children younger than 2 years old.

The incidence and repair rates for 1983 to 1995 were standardized per 10,000 live births. The magnitude and direction of a trend was determined using Pearson's correlation coefficient.

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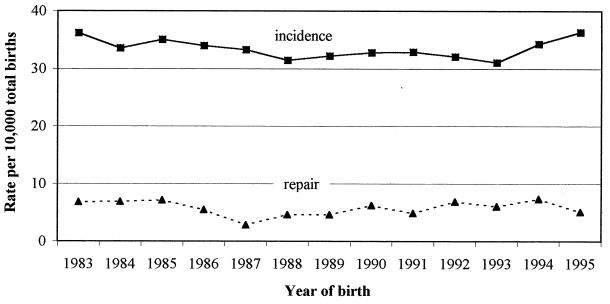

FIGURE 1. *Incidence and repair rates of hypospadias in NYS.*

RESULTS

A total of 11,938 hypospadias cases were reported in NYS during the specified 12-year period. During the same period, 2052 hypospadias cases were repaired. The standardized incidence and repair rates of hypospadias in NYS during this period are shown in Figure 1.

The incidence and repair rates of hypospadias in NYS did not change significantly during these 12 years ($r = -0.225$, $P = 0.45$ and $r = -0.010$, $P = 0.92$, respectively). In 1983, the standardized incidence and repair rates were 36.34/10,000 live births and 6.85/10,000 live births compared with 36.18/10,000 live births and 5.13/10,000 live births in 1995.

COMMENT

The results of this study found no trend in either incidence or surgical repair rates of hypospadias in NYS between 1983 and 1995 (Fig. 1). Pearson's correlation coefficient revealed an insignificant change, indicating random variability without an observable trend.

A recent data review of 29 registries, which monitor a total of 4 million births per year around the world, revealed a wide intercountry variation in the rates of hypospadias.[5] The variation was detectable in North America alone. The data from Atlanta, Georgia revealed a significant rise in incidence; Canadian data demonstrated a significant decrease; and the data from California showed no significant change. This wide variation in the incidence pattern of hypospadias is probably due to several factors.

First, the differences in the incidence of hypospadias may be due to environmental variations. Clinical studies have suggested that endocrine-disrupting environmental pollution may lead to the development of abnormal genitalia.[6–9] Clinical studies have shown that defects in testosterone receptors and testosterone metabolism by abnormal endocrine influences may lead to the development of abnormal male genitalia.[9] However, there is no clear evidence showing a direct relationship between the incidence of hypospadias and endocrine-disrupting chemical exposures.[10]

Second, the discrepancy in the incidence pattern of hypospadias may be due to different registry criteria. Distal or minor forms have generally been estimated to account for 70% of all cases.[3,11] The definition of minor forms, however, is unclear, because no clear anatomic marker that defines when "normal variation" stops and "minor forms" of hypospadias begins exists. Since most reported registry data do not indicate the severity of the hypospadias, it is possible that aggressive diagnosis of minor forms of hypospadias would lead to an overinflated incidence.

Third, the differences in the incidence of hypospadias may result from different registry methods. Currently, the methods to collect the data on congenital abnormalities are not unified. Some systems, such as the Hungarian Congenital Abnormality Registry, rely on physicians to report congenital abnormalities.[12] Others, like the nationwide Birth Defects Monitoring Program in the United States, tabulate diagnoses recorded on newborn discharge summaries.[13] Furthermore, some systems have gone through changes in registry methods. The Metropolitan Atlanta Congenital Defects Program (Birth Defects Monitoring Program) has shown an increasing trend in the incidence of hypospadias in the past. However, it changed its registry method, adopting a new birth defect coding system in 1985, which subsequently led to a decreased incidence of hypospadias being reported by their program.

As the data review of 29 registries and our study revealed, no absolute pattern can be detected in the incidence of hypospadias in the past 20 years. Each region and country had its own pattern of incidence, and the results from one region cannot be transposed to another region.

CONCLUSIONS

Epidemiologic studies in the United States and Europe found no consistent trend in the incidence of hypospadias. The results of the present study also revealed no statistically observable trend in either the incidence or surgical repair rates of hypospadias in NYS between 1983 and 1995. The variation in the incidence pattern of hypospadias is probably due to differences in several factors such as the environment, registry method, and diagnostic criteria.

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EDITORIAL COMMENT

Dr. Choi and colleagues have provided the practicing urologist with another piece of the complex puzzle that attempts to define the etiology of hypospadias. In this regard, unfortunately, there continues to be more questions than answers. The very tool that is used to determine hypospadias rates (ie, surveillance systems) may actually be a confounding variable in our attempt to determine hypospadias trends throughout the world. Surveillance systems are only as good as the documentation of anomalies reported in the medical records used by surveillance protocols. Although trends may be a means by which discoveries occur, they may also be an artifact of surveillance and inadvertently misguide. Using such systems, hypospadias rates have been reported to rise in Atlanta and Italy, remain the same in New York State and Canada, and decline in Spain. Clearly, a global trend has not been documented. What is apparent is that no identifiable cause for the regional trends, as reported here, has been determined nor ever been identified with certainty.

It may be easier to ask why hypospadias rates have been shown to increase in certain regions of the world. Is hypospadias the result of an environmental problem? No strong evidence exists proving such a link. Investigators have not demonstrated any seasonality, regional trends, known workplace exposure, or relation to industrialization. How about hormones, pesticides, pollutants, or "endocrine disruptors"? There is still no clear evidence. To refute a genetic link, evidence in twins discordant for hypospadias showed that the smaller twin was most likely to have hypospadias. Familial clustering of hypospadias may be polygenic, but also occurs as a feature of other syndromes with mendelian inheritance. So what is left? We can speculate that the factors that cause hypospadias must be related to maternal-fetal bloodflow, such that a vascular event prevents the delivery of "growth factors" to the embryo.

In attempting to explain the increased incidence of hypospadias in Atlanta, Paulozzi *et al.*[1] have disregarded the concept that we are diagnosing relatively more minor degrees of hypospadias. This is evidenced by the lack of a concomitant decline in the incidence of severe forms, which would be expected to be less prevalent if the minor cases were diagnosed relatively more often. Recently, our group in Atlanta has identified a greater than 10-fold increase in the incidence of hypospadias (11%) in small for gestational age (SGA) babies from 1996 to 1998. A trend toward a decreasing placental weight was also noted when SGA babies with hypospadias were compared with SGA babies without hypospadias. The degree of hypospadias did not correlate with prematurity or fetal weight.[2]

I believe that the "factors" resulting in SGA babies occur at a critical point in development, affecting both urethral and somatic growth. Although this may be overstating the obvious, hypospadias, like almost all birth defects, may be just one manifestation of a more global process.

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