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Case Report

Hypospadias in Twins

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ABSTRACT

Hypospadias is one of the most common birth defects among male children. Many associated risks for hypospadias have been reported like family history, older maternal age, nulliparity, high pre-pregnancy body mass index of mother, hypertension or preeclampsia, multiple gestations, low birth weight, and small for gestational age. In literature twinning as a risk factor has been consistently shown to be associated with hypospadias. We report a 2-year-old male child (one of the twins) who presented with proximal microphallic hypospadias.

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Introduction

Hypospadias is a common congenital disorder wherein the external urethral opening lies proximal to the normal glanular location [1, 2]. This defect is considered to be due to arrested development although embryos do not exhibit a similar appearing phase [3]. Hypospadias is known to occur 1 in every 300 males (0.3%). The risk of occurrence is nearly 13 times greater in first-degree relatives (brothers, fathers, offspring) [3]. Several studies have also reported regarding familial clustering of hypospadias with $\approx 7\%$ of the patients reporting hypospadias in first- or second-degree relatives [4, 5]. As reported by Brouwers *et al.*, low birth weight, being a twin or triplet, fertility treatments, mother being a diethylstilbestrol-daughter, paternal subfertility, obesity, prescriptive drug use, and familial occurrence of hypospadias were associated with increased risk of hypospadias in general [6]. Low birth weight (a proxy for placenta dysfunction) seemed much strongly associated with proximal hypospadias with an OR (95%CI) of 9.1 (3.4-24.2) as compared with distal and middle hypospadias at 2.6 (1.4-5.0) and 2.3 (0.8-6.5). We report a 2-year-old male child who presented with proximal microphallic hypospadias and has a twin brother who was operated at 9 months' age for perimembranous ventricular septal defect (L to R) with severe pulmonary arterial hypertension and sinus rhythm.

Case Report

A 2-year-old male child presented to the pediatric urological services with a small sized hypospadiac penis. He was one of the monozygotic twins (antenatal scans). The other twin was diagnosed to have perimembranous ventricular septal defect (L to R) associated with severe pulmonary arterial hypertension and sinus rhythm and underwent Sauvage patch closure of VSD at the age of 9 months. There was no history of consanguinity and the twins were born at full term. No history of use of drugs during pregnancy, assisted reproduction or, exposure to pesticides during pregnancy.

On examination the child was conscious and playful. The penis appeared microphallic with proximal penile hypospadias and chordee (Figure 1). The stretched penile length was 18mm and the diameter of glans was 11 mm. The testes were fully descended bilaterally, and no other obvious anomalies were seen. The parents were counselled regarding the starting of preoperative hormonal treatment with injection testosterone 2mg/Kg body weight once a month, for three months. The penile length and girth increased following injections and the parents were counselled for surgical repair of the hypospadias.

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Figure 1: a & b) Proximal penile hypospadias.

Discussion

There has been an increase in the rate of twins worldwide, probably because of assisted [5]. The rate of twin births in USA increased by 75% from ~ 18.9 to 33.2 twins per 1000 births) [7]. The reason for this increase has been attributed to increasing use of fertility treatments. It is well established that twins in comparison with singletons are at increased risk for several adverse outcomes including low birth weight, preterm birth and infant mortality [8]. Among the twins the monozygotic (MZ) twins are at an increased risk of birth defects compared to both singletons and dizygotic (DZ) twins [9].

The aetiology of isolated hypospadias is mainly multifactorial, and several factors such as genetic, endocrine, environmental, and clinical factors have all been observed. Twinning is a consistent risk factor that has been found to associated with hypospadias [10, 11]. Rider *et al.* reported that hypospadias occurred more often in twins compared with singletons (adjusted odds ratio (OR) 2.1 (95% CI: 1.5- 3.1) [9]. In birth weight discordant twins, hypospadias is more frequently observed in the twin with the lowest birth weight [12]. When genetic factors get ruled out in monozygotic twin pairs, Environmental etiological risk factors appear to be important. Visser *et al.* reported the incidence of hypospadias in monozygotic group was 4% (14/350) and dichorionic groups was 1% (3/303) ($p= 0.016$) respectively. In 11 of the 15 twin couples, hypospadias occurred in the twin with the lowest birth weight. The rate of hypospadias in twin infants small-for-gestational- age group was high (10%) as compared with 2% in the appropriate-for-gestational-age group ($p=0.002$). In a multivariate analysis the independently associated factors with hypospadias were monozygosity and small-for-gestational-age.

It is also well known that hypospadias occurs more frequently in small for gestation age (SGA) infants [13]. This association between SGA and hypospadias suggests that there could be an association between placental insufficiency and abnormal genital development. Nutrients and gonadotropins would become inadequate for the normal development of the two pairs of male gonads in twin pregnancies due to inadequate placentation or unequal placental sharing [14, 15]. The smaller twin would be more sensitive to relative deficit in hormones, hence at an increased risk of developing hypospadias. Monozygotic twins have an increased risk for hypospadias. Monozygotic and SGA were identified to be independently associated with hypospadias.

Funding

None.

Conflicts of Interest

None.

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