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CASE REPORT

Penile Duplication in Newborn with Multiple Anomalies

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We present a case of male, premature born child with multiple anomalies. The physical status was dominated by penile duplication, with additional, abortion scrotum in the median line, without palpable content and omphalocele with semi septum of the anus. By ultrasound examination of internal organs, we found a complex congenital heart anomaly, multiple anomalies in abdomen and micro calcifications in the central nervous system. General condition of the child required a prolonged mechanical ventilation and intensive care, so with clinical picture of liver failure at the age of 45 days occurred fetal outcome. **Key words: penile duplication, newborn.**

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1. INTRODUCTION

Penile duplication is rare anomaly with an incidence in 1 of 5.5 million live born (1). It can be presented alone or with associated anomalies. Schneider classified diphallus into three categories: duplication of the glans, bifid diphalia and complete penis duplication (2). Vilanov and Ravenous later added a fourth category: pseudodiphalia (3). Most authors agree that this is a defect connecting the genital tubercle. It is believed that diphallia in fetuses occurs between 23 and 25 days of gestation, when the injury, chemical agents or malfunctioned genome which deeply disturbed caudal cell mass of fetal mesoderm.

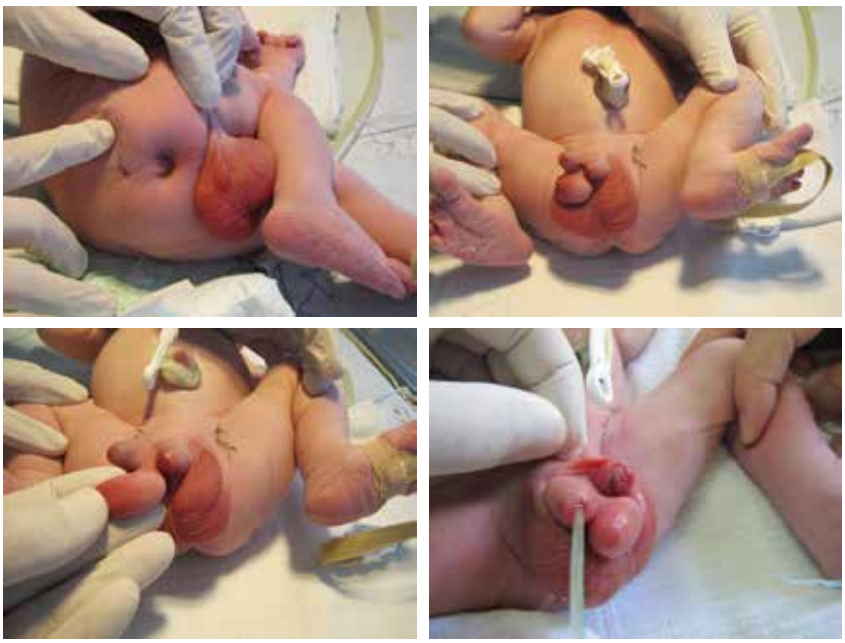
All patients with diphallia must be carefully evaluated with respect to the high incidence of associated anomalies of other organ systems. Prognosis and outcome depends largely on whether it is a case of solitary anomaly (which is less likely) or penile duplication with multiple associated anomalies. Treatment of the diphallus is based on surgical removal after careful evaluation of the anatomic relationships between related structures.

2. CASE REPORT

We present a case of male, premature born child hospitalized after birth at the neonatology department due to noticed anomalies of the genitals. This is the first child of young, phenotypically normal parents, with normal antenatal and family history, without medical supervision during pregnancy. Birth was completed naturally at the 34.5 week of gestation, with birth weight of 2560g, and birth length of 46cm. Apgar score in the first minute was assessed as 10. Physical examination found a penile duplication, with two completely separate, nearly equally developed penises, one with hypospadias and the other with normal meatus. Scrotum is well developed, with palpable testes and additional visible, abortion scrotum in the median line, with no palpable content. Visualized is omphalocele 4cm in diameter. Anus with prominent circular folds seems semi septed. Patients vital signs are stable and with normal initial laboratory test results, spontaneous meconial defecation, before the poor spontaneous voiding in both genitals. In the further course child does not tolerate enteral intake, secreted a yellow-green content in nasogastric probe and in projection of omphalocele. From the third day of life oliguria, then anuria, oxygen-dependent indirect support, the auscultation reveals audible precordial systolic noise intensity II-III/6 by the Levine. In laboratory tests gradually progresses mineral acid base imbalance with the elevation of parameters of renal and hepatic function. TORCH analysis was negative for IgM antibodies. Cariogram 46 XY.

ECHO examination of the heart shows the dominance of right heart cavities, discontinuity in the interatrial (10 mm) and interventricular septum (10 mm), with a single, common, atrioventricular regurgitate valve and one, exiting blood vessel from both ventricles.

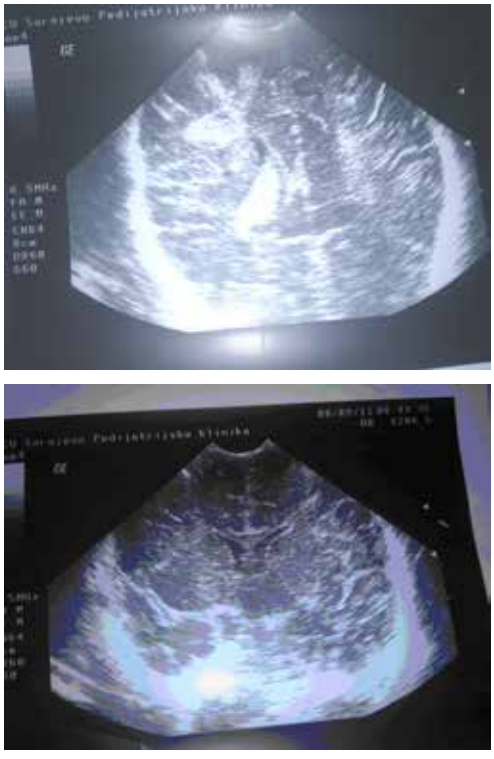
ECHO examination of the abdomen visualize multiple calcifications of the liver, kidney, and calcifications are present also interintestinally. Liver with coarse echo structure. Choledochal unclear monitored with a combination curved, non-echogenic, and tubular structures in its projection. Observed are three spleens (diameter 3.91, 3.95 and 2 cm). There is a duplicate of the urinary bladder, fully duplicated right ureter and right kidney channel system, with a probable vesicourethral reflux. Visible signs of the ductus omphaloentericus.



Figures 1-4. Penile duplication of newborn with multiple anomalies

Ultrasound of the CNS: show the existence of a regular chamber system. On both sides of the brain are present large hyperechogenic clearly defined zones, which correspond to irregular infarcts or focal accumulations of calcium. Given the static nature of the changes in the repeated findings it is less likely that these are hemorrhagic lesions.

Clinically, in the further course, the general aspect of a patient is poor, on mechanical ventilation, total parenteral nutrition, requiring daily correction of acidobase and mineral imbalance. The laboratory tests reveal liver insufficiency with elevation of transaminases and increased total bilirubin with the dominant direct fraction and despite the taken measures of intensive care, death occurs at the age of 45 days.



Figures 5 and 6. Results of Ultrasound examitaions

3. DISCUSSION

Diphallia is an extremely rare anomaly with only about 100 cases reported since the first described in 1609 by Johannes Jakob Wecker (4). Schneider classified diphallia into three groups: duplicate of the glans, bifid diphallia and complete diphallia (2), as we described in our patient, with a later added a fourth pseudodiphallia category (3). Meatus is described as hypospadic, epispadic or normal. In the presentation of six cases of diphallia Mirshemirania and colleague, three patients had a normal meatus, two hypospadia and one epyspadia. From the presented six patients, only one had a normal scrotum and the other five doubled (5). In our case of penile duplication, one had a hypospadic genital opening of the urethrae, and the other normal. Scrotum was also duplicated, one of which was well developed, with the testicles and the other abortive without palpable content, which coincides with the data of other authors and, where diphallia in most cases is associated with anomalies of the scrotal meatus. This anomaly is rarely occurs as a solitary, in most cases it is associated with other congenital anomalies.

In the cases described so far most common was the case of genitourinary and gastrointestinal tract anomalies, in the form of different variants within these duplicated systems (6-8). In our case it was a duplication of the blade, double right ureter and right kidney canal system, with the probable vesicourethral reflux, which was confined by cystogram. Also noted were polisplenia and complex cardiac anomalies (trunk arteriosus). With complete diphallia usually associated anomalies of the intestines (9), as in this case, where there was omphalocele with persistent ductus omphalomesentericus. With the available diagnostic methods, we found intestinal duplication, but by examining the clinical course and permanent control with ultrasound, it was most likely the case of the associated biliary atresia, with resultant liver failure.

There are multiple explanations for

Figure 7. New born after treatment

embryological diphalia and associated anomalies, summarized by Willson and Halowell (10, 11) but most authors agree that this is a defect connecting the genital tubercle. It is believed that diphallia in fetuses occurs between 23 and 25 days of gestation, when the injury, chemical agents or malfunctioning genome deeply disturbed fetal caudal cell mass of fetal mesoderm

All patients with diphallia must be carefully evaluated with respect to the high incidence of associated anomalies of other organ systems. Prognosis depends largely on whether it is a case of solitary anomaly (which is less likely) or penile duplication with associated anomalies. In our patient's poor general condition and the impossibility of surgical management of life-threatening congenital heart anomalies were crucial for an adverse outcome.

Treatment of the diphallus is based on surgical removal of the same, after careful evaluation of the anatomic relationships of related structures.

Conflict of interest: none decalred.

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