

Olgu Sunumu

Co-Occurrence Of Perineal Accessory Scrotum And Penoscrotal Transposition With Anal Atresia

Perineal Aksesuar Skrotum Ve Penoskrotal Transpozisyon İle Anal Atrezi Birlikteliği

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Abstract: A rare case of accessory scrotum and penoscrotal transposition with co-occurring anal atresia is reported because of its infrequency. Diagnosis, management and result of one-day-old newborn with high anorectal anomaly and perineal lipoma bearing accessory scrotum aimed to be presented in the light of literature screening.

Key words: congenital scrotal anomaly, accessory scrotum, penoscrotal transposition, anal atresia

Öz: Çok nadir görülmesi nedeni ile anal atrezinin eşlik ettiği aksesuar skrotum ve penoskrotal transpozisyon vakası sunulmuştur. Anorektal malformasyonu ve perineal lipom içeren aksesuar skrotumlu bir günlük yenidoğanın tanı ve yönetiminin literatür bilgisi ışığında sunumu amaçlanmıştır.

Anahtar sözcükler: konjenital skrotal anomali, aksesuar skrotum, anal atrezi

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1. INTRODUCTION

Congenital scrotal anomalies are rare anomalies. Penoscrotal transposition, ectopic scrotum, bifid scrotum, accessory scrotum are categories of congenital scrotal anomalies (1). Between these anomalies accessory scrotum and ectopic scrotum are extremely rare (2). Accessory scrotum (AS) presents like ectopic scrotal tissue in perineum or elsewhere without testicular tissue within it (3,4). AS can be observed as an isolated anomaly or with co-occurrence of other anomalies like genitourinary or anorectal anomalies (5). A rare case of accessory scrotum and penoscrotal transposition with concomitance of anal atresia is presented in this study.

2. CASE REPORT

A newborn boy with anal atresia and perineal accessory scrotum was referred to our department. In physical examination, there was anal atresia, bilaterally palpable high scrotal located testes and accessory scrotum

between penoscrotal transposed scrotum (Figure 1-2). Invertogram revealed a high anorectal anomaly. No other system pathologies were detected in multisystemic evaluation of the patient. Sigmoid diverting ostomy was created as a first step of repair on postnatal first day. MRI was planned for obtaining borders of swelling within accessory scrotum. MRI images of perineum reported a 5x2.5 cm lipomatous lesion within the accessory scrotum extending through left scrotum. Rectovesical fistula was detected in distal colon X-ray before definitive operation. At the postnatal twelfth month rectovesical fistula was dissected and separated from the bladder wall and anastomosis was accomplished successfully to the perineum. After recovery of definitive operation of anal atresia, perineal lipoma and accessory scrotum excision was accomplished on postnatal thirteenth month (Figure 3-4). Pathology of excision material was reported as lipoma. Colostomy closure is planned on postoperative sixth month after definitive operation. Patient is doing well after operations till this day.



Figure-1: Accessory scrotum, penoscrotal transposition

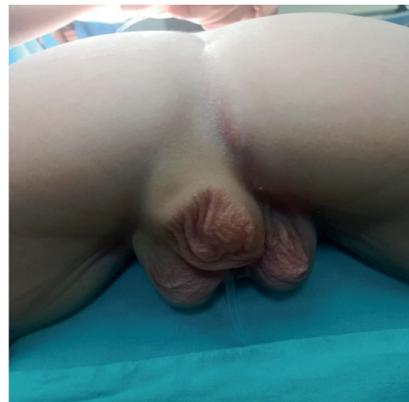


Figure-2: Associated anal atresia

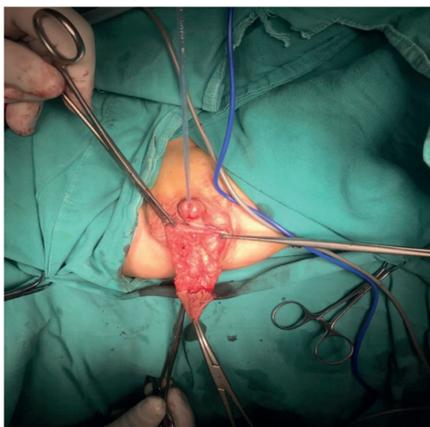


Figure-3: Excision of accessory scrotum and perineal lipoma



Figure-4: Postoperative look after accessory scrotum and lipoma excision.

3. DISCUSSION

The scrotum is formed by migration and the fusion of labioscrotal swellings in the midline. Fourth week of gestation is the time period for appearance of labioscrotal swellings and after twelfth week of gestation migration to the caudal portion occurs. Lack in the migration and division of the labioscrotal swellings are the probable causes of congenital scrotal anomaly (1). Lamm and Kaplan suggested that early division or abnormal migration of labioscrotal swelling can cause scrotal anomalies (6). Accessory scrotum is a less frequent congenital scrotal anomaly (5,7). First case of accessory scrotum was reported in 1930 by Kimura (8). This anomaly is characterized by additional scrotal tissue and normally developed scrotum with testes. There are few embryological explanations about scrotal anomaly's developmental etiology. Sule hypothesized development of AS to be consequence of disruption the continuity of caudal labioscrotal swelling by intervening mesenchymal tissue (9). Another hypothesis developed by Takayasu accuses early division and teratoid growth of pluripotent labioscrotal tissue elements in development AS (10).

Despite the advances in prenatal screening, perineal masses are mostly detected postnatally (11). There are only three cases detected prenatally. (11,12,13). Antenatal detection of perineal mass requires careful evaluation of all systems for any associated congenital anomalies (14). No antenatally detected anomalies reported in our patient.

High incidence (%72.5) of co-occurring subcutaneous tumors with AS reported in the literature (5,11). Lipomas are very rare in neonates but most cases co-occurring with AS are reported to be lipoma, followed by hamartoma and lipoblastoma (5,11). As a result of radiological and pathological evaluations subcutaneous tumor within accessory scrotum reported as lipoma in our case.

Beside subcutaneous tumors, genitourinary (%35.2), skeletal (%4.7) and anorectal anomalies (%18.6) accompany AS (5). High type of anorectal malformation was detected in our patient. No other genitourinary and skeletal anomalies were detected.

We experienced a very rare case of AS accompanied with high anorectal malformation. Difference of our case from other cases reviewed in the literature is togetherness of two congenital anomalies like accessory scrotum and penoscrotal transposition in the same patient. Another reason for presentation of this case is rareness of anal atresia concomitance. It is important for clinician to be aware of co-occurrence of other system anomalies in the presence of antenatally detected perineal mass. Although removing of perineal mass and reconstruction of accessory scrotum do not require difficult approaches, accompanied

anomalies can make this anomaly more challenging. Primary goal in management of this case was repair of anal atresia and rectovesical fistula. We advice rational evaluation of patient born with perineal lipoma and step by step planning of anomaly repair according to urgency.

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