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Case Report

Unusual association of ectopic scrotum with anal stenosis: a case report

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ABSTRACT

Congenital anomalies of the scrotum are rare and include penoscrotal transposition, bifid scrotum, ectopic scrotum, and accessory scrotum. Suprainguinal ectopic scrotum is characterized by the abnormal location of a hemiscrotum outside its usual position and is frequently associated with other congenital anomalies. We report a male infant with left suprainguinal ectopic scrotum associated with anal stenosis, an association not previously described. A term male born by repeat caesarean section presented with a left hemiscrotum located in the ipsilateral inguinal region, harboring a palpable testis. Anal stenosis and a sacrococcygeal pit were also noted. Work-up included lumbosacral MRI, which excluded spinal cord anomalies. The patient underwent a sigmoid colostomy, followed by a limited posterior sagittal anorectoplasty, and subsequently underwent colostomy closure. At 18 months, one-stage scrotoplasty with a random-pattern transposition flap and left orchidopexy were performed jointly by the pediatric urology and plastic surgery teams. Postoperative follow-up at 21 months demonstrated an orthotopic, well-vascularized scrotum with viable testes. Ectopic scrotum results from defective development of the gubernaculum and labioscrotal swellings. Its concurrence with anorectal malformations suggests a broader mesodermal defect. Early, individualized, multidisciplinary management is essential. Our case is, to our knowledge, the first report of ectopic scrotum associated with isolated anal stenosis. Timely diagnosis and coordinated surgical management can restore normal anatomy and function with excellent aesthetic outcomes. Awareness of possible associated anomalies is crucial for comprehensive evaluation.

Keywords: Ectopic scrotum, Congenital anomalies, Scrotoplasty, Orchidopexy, Anal stenosis

INTRODUCTION

Congenital anomalies of the scrotum are an infrequent condition that are classified into penoscrotal transposition, bifid scrotum, ectopic scrotum, and accessory scrotum.[1] Within this subdivision, ectopic scrotum represents a rare form, characterized by the abnormal location of one of the hemiscrota outside its usual position. It may be located in the inguinal, suprainguinal, infrainguinal, or perineal region, with the suprainguinal location being the most frequent.[2,4]

The embryological development of the scrotum begins between the fourth and sixth weeks of gestation with the appearance of the labioscrotal swellings. Between weeks 12 and 14 of gestation, these swellings fuse at the midline and migrate inferomedially to form the scrotum. Testicular descent occurs later, between weeks 26 and 35 of gestation, once the gubernaculum has fused with the labioscrotal swellings. A defect in the development of the gubernaculum can alter this process, preventing the proper fusion and positioning of the scrotum.[2]

Ectopic scrotum is usually associated with other congenital malformations; approximately 70% of suprainguinal ectopic scrotum cases are related to ipsilateral anomalies, such as renal dysplasia, renal agenesis, ectopic ureter, testicular atrophy, penile torsion, or even diphallia.[1] In these cases, the testis may be either present or absent within the ectopic scrotum.[5]

The treatment of ectopic scrotum is surgical, and different strategies exist depending on the complexity of the case. Among the most common options are one-stage repair, which involves simultaneous scrotoplasty and orchidopexy, and two-stage repair, which may consist of scrotoplasty followed by orchidopexy or the reverse.[3]

This case report aims to describe the clinical presentation and surgical management of a patient with left ectopic scrotum associated with an anorectal malformation, highlighting its embryological relevance and the importance of a multidisciplinary approach.

CASE REPORT

Male patient, product of a term repeats caesarean section, without prenatal or perinatal complications. At birth, left hemiscrotal ectopia was identified, with the ectopic hemiscrotum located in the ipsilateral inguinal region (Figure 1).

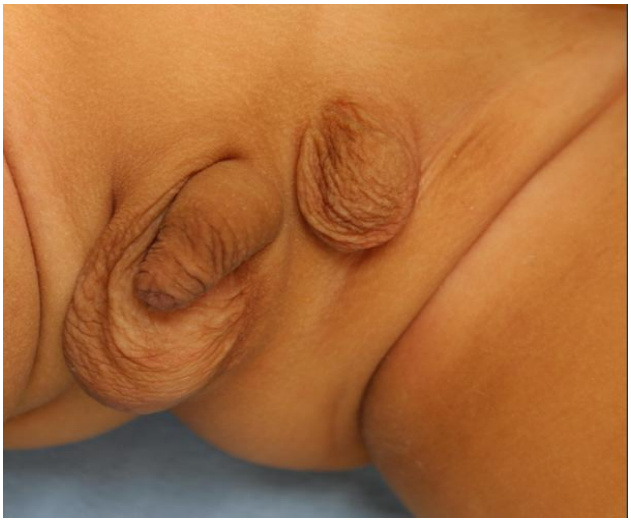

Figure 1: Left hemiscrotal ectopia was identified, with the ectopic hemiscrotum located in the ipsilateral inguinal region.

At 6 months of age, he attended a paediatric urology consultation for evaluation. On physical examination, the left hemiscrotum was located in the inguinal region, with a palpable left testis inside. The right hemiscrotum and testis were normal. Additionally, the anus was identified within the muscular complex; rectal examination was not possible due to stenosis, as the corresponding Hegar dilator for his age could not be passed-only a size seven dilator was able to pass (Figure 2). A sacrococcygeal dimple was identified in the sacral region. The mother reported daily pasty stools.

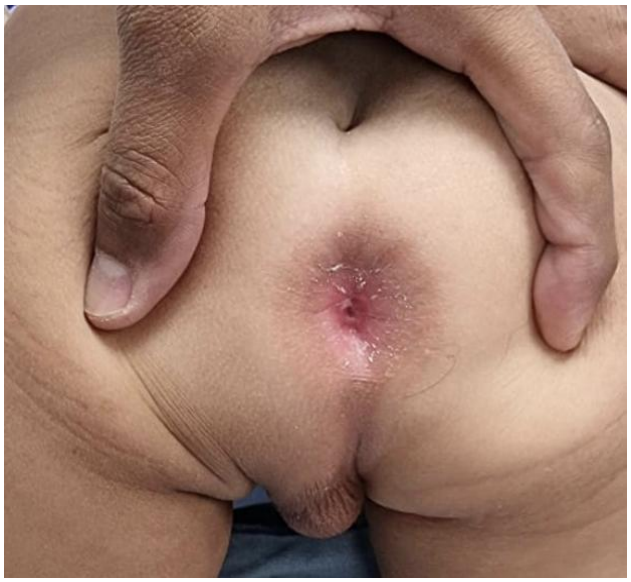

Figure 2: The anus was identified within the muscular complex.

As part of the diagnostic work-up, a lumbosacral magnetic resonance imaging (MRI) was performed, which identified the presence of a sacrococcygeal dimple. The case was evaluated by the neurosurgery department, which concluded that there was no indication for surgery and that associated spinal cord malformations were ruled out. A contrast enema was performed, revealing a dilated colon and redundant sigmoid colon, leading to the diagnosis of rectal stenosis (Figure 3).

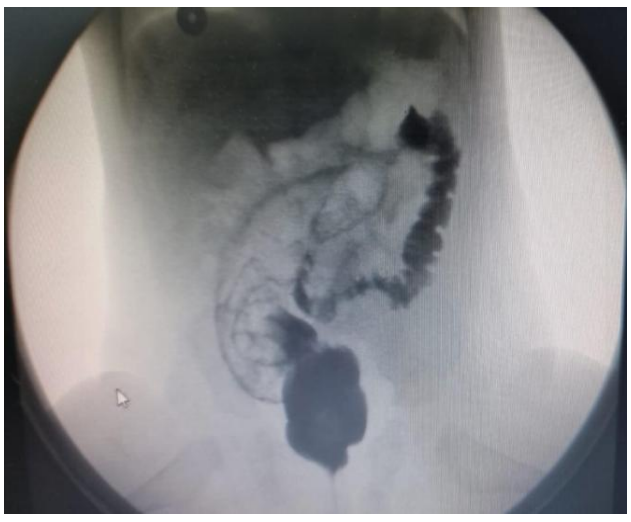

Figure 3: Contrast enema revealing a dilated colon and redundant sigmoid colon, leading to the diagnosis of rectal stenosis.

A sigmoid colostomy was performed, followed by a limited posterior sagittal anorectoplasty, without surgical

complications. Later, the sigmoidostomy was closed (Figure 4).

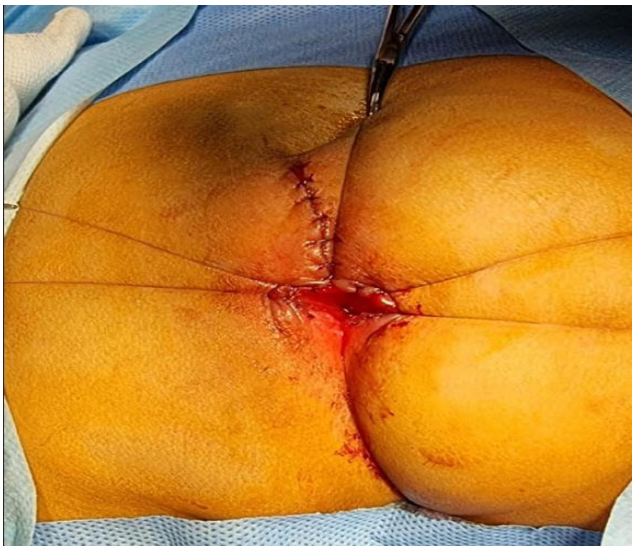

Figure 4: Limited posterior sagittal anorectoplasty.

A joint approach was taken by the pediatric urology and plastic surgery teams, with the scrotoplasty and left orchidopexy scheduled. The technique consisted of a 3:1 cutaneous transposition flap with a random vascular pattern (Figure 5).

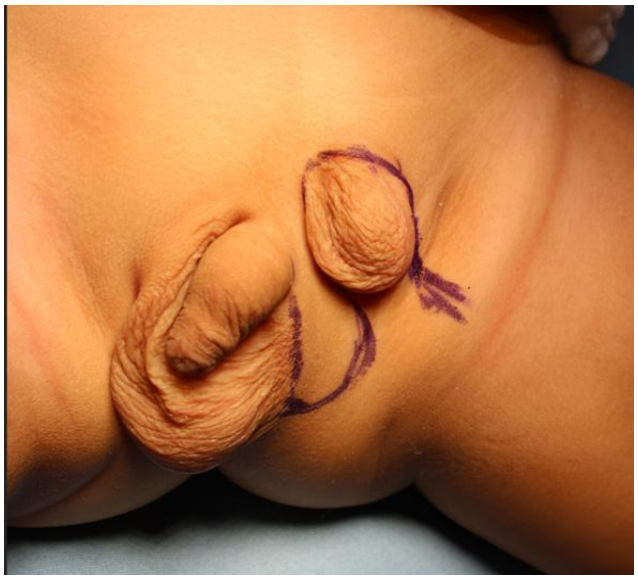

Figure 5: Preoperative surgical marking with the cutaneous transposition flap along with the scrotoplasty and left orchidopexy.

Dissection was performed over the subcutaneous tissue, preserving the vascularity of the subdermal plexuses. The flap was raised from the donor site and rotated on its base.

Simultaneously, the subcutaneous plane beneath the ectopic testis was dissected to create the defect at the recipient site. Both the cutaneous flap and the testis were transposed to the scrotal region. Layered closure was performed using inverted simple stitches and subdermal closure with 5-0 monocryl. Finally, the left testis was fixed to the scrotal sac in an anatomical position, along with the contralateral testis, using the same closure technique. At follow-up at 1 year and 9 months of age, the patient presented with the left hemiscrotum in an orthotopic location, containing the left testis, which was of adequate size and vascularity (Figure 6).

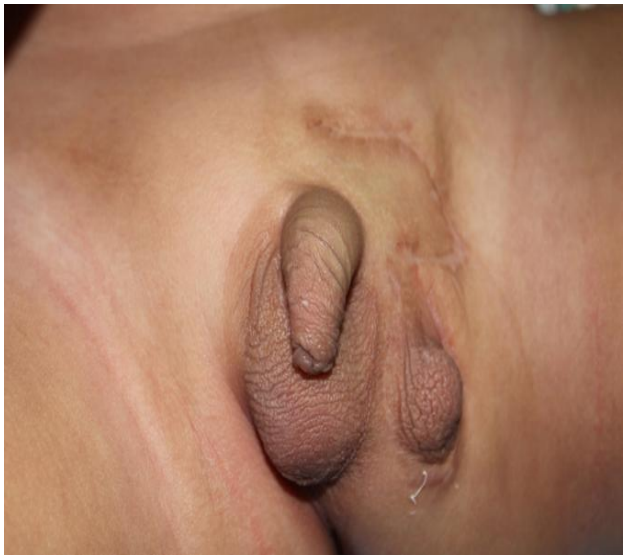

Figure 6: Follow-up at 1 year and 9 months of age, left hemiscrotum and testicle in an orthotopic location.

DISCUSSION

Ectopic scrotum represents an uncommon anomaly of the embryological development of the genitourinary system. Its clinical presentation may vary depending on the degree of ectopia, its location, and the associated malformations. In the described case, a left suprainguinal ectopic scrotum was observed, which is consistent with the most frequently reported location in the literature.[2,4] The presence of the testis within the ectopic hemiscrotum was also noted, a finding that has been previously documented. In some cases, the testis may be absent or located intra-abdominally or in the inguinal region.[1,5]

From an embryological perspective, this malformation is associated with alterations in the development of the gubernaculum and its interaction with the labioscrotal swellings.[2] A defect in this process explains the ectopic scrotum and failed testicular descent.

Along with the scrotal and testicular ectopia, the patient presented a congenital malformation of the lower gastrointestinal tract and a sacrococcygeal dimple without spinal cord abnormalities, which shows how the coexistence of multiple malformations in the same patient requires a comprehensive multidisciplinary approach. This approach should include diagnostic and imaging studies to

enable evaluation by specialized surgical teams, thereby achieving an adequate functional and aesthetic outcome.

In this particular case, the simultaneous presence of a scrotal malformation, anorectal malformation, and a finding in the spinal region made it appropriate to rule out the VACTERL association (vertebral, anorectal, cardiac, tracheoesophageal, renal, and limb anomalies). To establish the diagnosis of this association, the presence of at least three of these six anomalies is required. Its incidence is estimated to range from 1 in 10,000 to 1 in 40,000 live births.[10]

The surgical treatment of ectopic scrotum aims to restore the functional and aesthetic anatomy of the scrotum, as well as to ensure proper fixation of the testis to preserve its endocrine and spermatogenic function. In this case, a one-stage scrotoplasty and orchidopexy was chosen, which is indicated when there are no significant anatomical limitations, and the viability of the flap allows for proper reconstruction.[3] The technique, which utilized a transposition flap and a random vascular pattern, proved effective, yielding a satisfactory anatomical and functional result during follow-up.

Clinical follow-up confirmed an uneventful postoperative course, with the left hemiscrotum and testis in an orthotopic location, of normal size and vascularity. This supports the importance of early and well-planned intervention to prevent long-term complications such as infertility, testicular atrophy, or psychological impact due to alterations in genital morphology.

The relevance of this case report lies in the unusual association between ectopic scrotum and anal stenosis; the coexistence of these two malformations has not been previously documented in the medical literature. Ectopic scrotum may present alongside other congenital anomalies, such as complex genitourinary or anorectal malformations; however, the specific coexistence with anal stenosis represents an unusual clinical finding.

CONCLUSION

Ectopic scrotum is a rare congenital anomaly whose timely diagnosis is essential to avoid long-term functional complications. Its clinical presentation may vary and is frequently associated with other congenital malformations, as occurred in the presented case, in which it coexisted with an anorectal malformation.

This highlights the importance of a comprehensive and multidisciplinary evaluation, from initial diagnosis to implementing an integrated approach for addressing the congenital malformations present in the patient.

Proper surgical planning allows not only the restoration of the functionality of the involved structures-as in the case of the testes, where viability was preserved-but also the achievement of an adequate aesthetic appearance. The one-stage scrotoplasty with transposition flap and orchidopexy, performed jointly by pediatric urology and plastic surgery, proved to be effective, providing satisfactory outcomes at medium-term follow-up.

This case highlights the importance of an individualized, multidisciplinary, and early approach to managing congenital anomalies of the scrotum, as well as the necessity of postoperative follow-up to ensure the patient's proper development and verify the functionality of the structures.

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