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Clinical case

Strangulated inguino-labial hernia associated with hermaphrodism in a child

Hernie inguino-labiale étranglée associée à l'hermaphrodisme chez l'enfant

Y Sow*[1], I Diallo[1], MS Bangoura[2], K Kamano[1], AA Bah[1], AT Sow[1], IM Diallo[1], LT Soumaoro[2]

Résumé

Introduction : L'objectif de ce travail était de décrire les aspects cliniques et thérapeutiques des hernies inguino-labiales chez les patients présentant un trouble du développement sexuel.

Cas clinique : Il s'agit d'une patiente âgée de 4 ans présentant une tuméfaction inguino-labiale gauche congénitale, douloureuse et irréductible, dans un contexte de trouble du développement sexuel. Un bilan préopératoire a été réalisé. Après induction de l'anesthésie générale, la hernie s'est réduite spontanément.

L'exploration peropératoire a retrouvé un sac herniaire contenant l'utérus et les ovaires, adhérant au sac. Le geste chirurgical a consisté en une libération prudente des structures, une réintégration des organes, suivie d'une fermeture du sac herniaire et d'un renforcement pariétal.

Les suites opératoires ont été simples. Aucun événement indésirable n'a été noté après un suivi de 1, 3 et 6 mois.

Conclusion : La hernie inguino-labiale étranglée chez l'enfant présentant un trouble du développement sexuel est une entité rare. Le diagnostic repose sur des arguments cliniques et paracliniques. La prise en charge est chirurgicale et nécessite une approche multidisciplinaire. Le dépistage précoce pourrait améliorer le diagnostic dans des contextes socioculturels où ces affections restent stigmatisées.

Mots-clés : hernie inguino-labiale, étranglement, trouble du développement sexuel, prise en charge, Guinée.

Abstract

Introduction: The aim of this study was to describe the clinical and therapeutic features of inguinolabial hernias in patients with disorders of sex development.

Clinical case: A 4-year-old patient presented with a congenital, painful, and irreducible left inguinolabial swelling in the context of a disorder of sex development. A preoperative assessment was performed. Following induction of general anesthesia, spontaneous reduction of the hernia occurred.

Intraoperative findings revealed a hernial sac

containing the uterus and ovaries, adherent to the sac. Surgical management consisted of careful dissection, reduction of the contents, hernia sac closure, and abdominal wall reinforcement. The postoperative course was uneventful. No complications were observed at 1, 3, and 6 months of follow-up.

Conclusion: Strangulated inguinolabial hernia in children with disorders of sex development is rare. Diagnosis is based on clinical and paraclinical findings. Management is surgical and requires a multidisciplinary approach. Early detection may improve diagnosis in sociocultural contexts where such conditions are stigmatized.

Keywords: inguinolabial hernia, strangulated, disorder of sex development, management, Guinea.

Introduction

Inguinal hernias are a common condition in the pediatric population. In girls, they may correspond to ovarian hernias, defined as protrusions through the inguinal canal above the inguinal ligament (1).

Strangulation represents the most serious complication and constitutes a true diagnostic and therapeutic emergency (2). These hernias are typically inguino-scrotal in males and inguino-labial in females.

Associations with other congenital anomalies are not uncommon. Disorders of sex development (DSD) encompass a heterogeneous group of anatomical and clinical conditions, including gonadal dysgenesis and ovotesticular DSD (3).

Sexual ambiguity encompasses a wide spectrum of anatomoclinical presentations, including gonadal dysgenesis, pseudohermaphroditism, and true hermaphroditism, as reported by Mansouri F et al. in Morocco (3).

Several case reports have highlighted the association between disorders of sex development and inguinal hernias. Ceylan K et al. in Türkiye described a 21-year-old patient presenting with cryptorchidism and a left inguinal mass, in whom internal genital organs were identified during surgery (4). Similarly, Fukui S et al. in Japan reported the case of a three-month-old boy with an impalpable right testis, left inguinal hernia, and penoscrotal hypospadias with asymmetry of the external genitalia; intraoperatively, female internal genital organs were found within the hernial sac (5).

The rarity of this condition and the limited number of reported cases justify the present study.

Clinical case

A 4-year-old patient presented with a congenital, painful, and irreducible left inguino-labial swelling in the context of a disorder of sex development.

A preoperative assessment was performed. Following induction of general anesthesia, spontaneous reduction of the hernia was observed.

Surgical exploration revealed a hernia sac containing the uterus and ovaries, which were adherent to the sac. The intestinal loops were viable.

The surgical procedure consisted of careful dissection, reduction of the hernia contents, closure of the hernia sac, and reinforcement of the abdominal wall.

Management took into account the disorder of sex development and involved consultation with a pediatric surgeon.

The postoperative course was uneventful. The length of hospital stay was 7 days. No complications were observed at 1, 3, and 6 months of follow-up.

Figure 1: Clinical view of a left inguino-labial swelling in a 4-year-old patient, consistent with an inguinal hernia in the context of a disorder of sex development.

Figure 2: Congenital left inguinolabial swelling in a 4-year-old child, presenting as a tense, rounded, irreducible mass, suggestive of a strangulated inguinolabial hernia in the context of a disorder of sex development.

Discussion

- *True hermaphroditism and disorders of sex development*

True hermaphroditism represents one of the rarest forms of disorders of sex development (DSD), accounting for approximately 5% of cases (6). It is also referred to as ovotesticular DSD and constitutes a rare intersex condition.

In most reported cases (approximately 90%), patients present a 46,XX karyotype. Less frequently, 46,XX/46,XY mosaicism may be observed, while rare cases with a 46,XY karyotype have also been documented (6).

- *Inguino-labial hernia associated with disorders of sex development*

Ceylan K et al. described true hermaphroditism as a heterogeneous condition with both phenotypic and genetic variability. Gonadal tissue may be located along any point of the embryonic testicular descent pathway and is frequently associated with inguinal hernia (4).

Similarly, Gujar et al. reported a case of a 30-year-old man with unilateral right cryptorchidism and a left obstructed inguinal hernia containing a uterus and a fallopian tube. This condition, identified intraoperatively, corresponded to an inguinal uterine hernia, a male type I form of persistent Müllerian duct syndrome (7).

According to Gujar NN et al., persistent Müllerian duct syndrome is a rare form of male pseudohermaphroditism characterized by the persistence of Müllerian duct derivatives in otherwise phenotypically and genotypically normal males (7).

- *Surgical management*

Gujar et al. reported a surgical approach consisting of right orchidopexy. The left testis and remnants of the female genital tract were excised. Histopathological examination revealed germinal aplasia of the testicular tissue. Karyotype analysis could not be performed due to technical limitations (8–10).

In a similar report, Fukui S et al. performed herniorrhaphy associated with excision of the right gonad, uterus, and fallopian tubes. Intraoperative exploration of the left gonad revealed an ovotestis. The ovarian component was excised, and a left testicular biopsy was performed. A one-stage hypospadias repair using the Koyanagi technique was also carried out. Histological findings demonstrated ovarian stroma in the right gonad and left ovary, while only Sertoli cells were identified in the left testicular biopsy specimen (5).

Conclusion

Strangulated inguino-labial hernia in children with disorders of sex development is a rare condition.

Diagnosis is based on clinical and paraclinical findings, and management is primarily surgical, requiring a multidisciplinary approach.

Early detection may improve diagnosis, particularly in sociocultural contexts where such conditions remain stigmatized.

***Correspondence :**

SOW Youssouf

sowyoussouf3@gmail.com

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1 : Pita Prefectural Hospital
2 : Conakry University Hospital Center

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