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Understanding Crossed Testicular Ectopia: An Embryological Perspective Through Case Presentation

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Abstract: ***Background: Crossed testicular ectopia (CTE), or transverse testicular ectopia, is a rare congenital anomaly in which both testes descend into the same hemiscrotum. It is frequently associated with inguinal hernia, persistent Müllerian duct syndrome (PMDS), hypospadias, and other genitourinary abnormalities. Fewer than 150 cases have been reported worldwide, making it an important subject for embryological and clinical study. Aim: To present two cases of CTE and discuss the embryological perspectives and clinical implications, highlighting the importance of early recognition and surgical management. Methods: Two infants presenting with an ipsilateral inguinal hernia and a contralateral non-palpable testis were evaluated. Surgical exploration confirmed that both testes were located in the same inguinal canal, with preservation of normal spermatic cord origin. Herniotomy and orchiopexy were performed. A literature review was conducted to analyse embryological theories and classification. Results: Both cases demonstrated classical features of CTE. No Müllerian remnants were identified. Surgical outcomes were favourable, with successful orchiopexy and hernia repair. Review of literature revealed multiple embryological hypotheses, including Wolffian duct adhesion, aberrant gubernacular development, defective inguinal ring formation, and the mechanical influence of Müllerian remnants. CTE is classified into three types: Type I (hernia alone), Type II (associated with Müllerian remnants), and Type III (associated with other genitourinary anomalies). Conclusion: CTE exemplifies the complex interplay between embryological development and clinical manifestation. Early recognition and timely surgical intervention are essential to prevent complications such as infertility, malignancy, and recurrent hernia. Case-based reporting enriches understanding of its pathogenesis and guides operative strategies.***

Keywords: Crossed testicular ectopia; Transverse testicular ectopia; Persistent Müllerian duct syndrome; Inguinal hernia; Gubernaculum; Wolffian duct adhesion; Testicular descent anomaly; Orchiopexy; Embryology; Genitourinary malformations.

1. Introduction

Crossed testicular ectopia (CTE), also known as transverse testicular ectopia, is a rare congenital anomaly in which both testes migrate to the same hemiscrotum. It is often associated with conditions such as persistent Müllerian duct syndrome (PMDS), inguinal hernia, hypospadias, pseudo-hermaphroditism, and scrotal abnormalities. [1-3]

The anomaly was first described by Von Lenhossek in 1886[1], and since then, fewer than 150 cases have been reported worldwide, highlighting its rarity and clinical significance. The condition typically presents in early childhood, most often during surgical exploration for an inguinal hernia or undescended testis. Clinically, it manifests as an ipsilateral inguinal hernia with a contralateral non-palpable testis, and diagnosis is usually confirmed intraoperatively.

From an embryological perspective, CTE provides a unique window into the complex process of testicular descent. Normally, the testes descend from the abdominal cavity into the scrotum under the guidance of the gubernaculum and hormonal influences. In CTE, this pathway is disrupted, leading to aberrant migration of one testis across the midline. Several theories have been proposed to explain this anomaly, including abnormal gubernacular development, persistence of Müllerian structures, and early fusion of Wolffian ducts.

Understanding CTE is important not only for its embryological implications but also for its clinical consequences. Early recognition and surgical management are essential to prevent complications such as infertility, testicular malignancy, and recurrent hernia. Case-based presentations of CTE therefore serve as valuable educational tools, bridging embryological theory with practical surgical outcomes.

2. Methodology

Case Description: We present two cases of CTE, where the ectopic testis maintained its normal spermatic cord origin from the appropriate side. In both cases, the testes were located in the inguinal region with an empty contralateral hemiscrotum, typically identified around 4 months of age [4-6].

Embryological Insights

1) Abnormal Testicular Descent Pathways

- Normally, descent is guided by the gubernaculum and hormonal signals (INSL3, testosterone).
- In CTE, misdirected gubernacular attachment may cause one testis to follow the contralateral inguinal canal.
- This suggests a mechanical misrouting rather than intrinsic gonadal abnormality [2].

2) Hormonal Influence and INSL3/Testosterone Defects

- Insulin-like peptide 3 (INSL3) secreted by Leydig cells is crucial for transabdominal descent.

- Defects in INSL3 signalling or androgen receptor pathways may predispose to abnormal migration.
- This highlights a molecular basis beyond structural anomalies[5].

3) Müllerian Duct Remnants and PMDS
- Persistent Müllerian duct syndrome (PMDS) is frequently associated with CTE.
- Müllerian structures (uterus-like tissue, fallopian remnants) can act as mechanical tethers, pulling one testis across the midline.
- This explains why CTE is often seen with hernia sacs containing Müllerian remnants[3].

4) Abnormal Development of Mesonephric (Wolffian) Ducts
- Fusion or adhesion of Wolffian ducts during early embryogenesis may cause testes to migrate together.
- This theory explains cases where both spermatic cords remain anatomically normal but converge in one inguinal canal[2,3].

5) Defective Inguinal Canal Formation
- Maldevelopment of the internal inguinal ring may allow one testis to cross over.
- This structural defect could explain why CTE is often associated with inguinal hernia[5].

6) Genetic and Developmental Factors
- Rare familial cases suggest a possible genetic predisposition.
- Mutations affecting sex differentiation genes (e.g., SOX9, WT1, SF1) may contribute to abnormal descent.
- However, no single gene has been definitively linked to CTE[7-10].

7) Embryonic Positioning Hypothesis
- Some authors propose that early misplacement of the gonadal ridge during embryogenesis may predispose one testis to cross the midline.
- This theory emphasizes positional errors at the earliest stages of gonadal development.

3. Discussion

Crossed testicular ectopia (CTE) remains a rare but intriguing anomaly of testicular descent. Clinically, the most common presentation is indeed an inguinal hernia with an ipsilateral palpable testis and a contralateral non-palpable testis. The diagnosis is often incidental, made during herniotomy or exploration for undescended testis, although advances in imaging have allowed for occasional preoperative identification. Ultrasonography and MRI can sometimes demonstrate both testes within the same inguinal canal, but their sensitivity remains limited, making intraoperative discovery the norm[4-6].

Classification and Clinical Associations
The classification into three types- hernia alone (Type I), association with Müllerian remnants (Type II), and association with other genitourinary anomalies (Type III)—is clinically useful. It not only guides surgical planning but also alerts clinicians to possible syndromic associations. Type II, linked with persistent Müllerian duct syndrome (PMDS), is particularly significant because Müllerian remnants may complicate orchiopexy and raise questions about fertility and malignancy risk[3,5].

Etiological Theories Beyond Berg and Kimura
- Berg's hypothesis: Both testes developing from the same genital ridge suggests a fundamental embryological misplacement.[2]
- Kimura's classification: Differentiating unilateral versus bilateral vas deferens origin provides anatomical evidence for whether the ectopia is due to unilateral origin or true migration [3].
- Mechanical theories: Most authors emphasize mechanical causes- abnormal gubernacular development, Wolffian duct adhesion, or defective inguinal ring formation—as the most plausible explanations [2,5].
- Hormonal and molecular factors: More recent insights suggest that defects in INSL3 or androgen receptor signaling could disrupt normal descent, adding a molecular dimension to the mechanical theories [5].
- Genetic predisposition: Rare familial cases hint at possible genetic contributions, though no single gene has been definitively implicated[7-10].

Clinical Implications
The importance of recognizing CTE lies in its potential complications:
- Infertility: Malpositioned testes are at risk of impaired spermatogenesis.
- Malignancy: Ectopic testes carry a higher risk of germ cell tumors, particularly if diagnosis and orchiopexy are delayed.
- Hernia recurrence: Inguinal hernia is almost universally associated, and failure to recognize CTE may lead to incomplete repair[4,10].

Surgical Considerations
Management typically involves herniotomy and orchiopexy. In Type II cases, careful excision or preservation of Müllerian remnants may be required, depending on their extent and impact on fertility. Preservation of vascular supply is paramount, as both testes may share close anatomical relationships [4-6].

Integrative Perspective
Taken together, CTE exemplifies the interplay of embryological misdirection and clinical manifestation. While mechanical theories remain dominant, emerging molecular and genetic insights suggest a multifactorial etiology. Each case report adds valuable evidence, refining our understanding of this rare anomaly and guiding optimal management strategies [3-5].

4. Conclusion

Crossed testicular ectopia (CTE) is a rare congenital anomaly that continues to challenge both embryologists and clinicians [1–3,7–10]. Early recognition and timely surgical management are essential to prevent complications such as infertility, malignancy, and recurrent hernia [4–6,8–10]. Case-based reporting remains invaluable, as each

documented instance contributes to refining embryological theories and guiding operative strategies [7–10].

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