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A Rare Presentation of Penopubic Testis in an Infant

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ABSTRACT

An ectopic testis is defined as one that has emerged from the external ring but fails to reach the scrotum. There are, in general, 5 sites of ectopic testis. A rare case of penopubic ectopic testis is reported here. A 6-month-old infant presented with penopubic ectopic testis. An orchidopexy was performed. During the operation, a gubernacular attachment was not found on the testis. Although Lockwood's 5 tails of gubernaculum form a commonly quoted mechanism of ectopic testis, our case could not fit in this mechanism since the gubernaculum was absent. A break in the gubernaculum during testicular descent, as suggested by Sonneland, leaving the testis radar-less after its exit from the superficial ring, may be an explanation for ectopic location in our case.

INTRODUCTION

An empty scrotum may indicate a retractile testis, undescended testis, testicular agenesis, or an ectopic testis. By definition, an ectopic testis is one that has emerged from the superficial inguinal ring but fails to reach the scrotum [1]. The described sites for ectopic testis are: within the superficial inguinal pouch, femoral, perineal, transverse (contralateral hemiscrotum), and penopubic, in descending order [2]. The etiology of ectopia seems unclear, with many explanations in the literature. We report an unusual ectopic testis at the penopubic site, where the gubernaculum was absent but sufficient length of the vessels was there to allow tension-free orchidopexy in the scrotal sac.

CASE REPORT

A 6-month-old infant presented with suprapubic swelling since birth, and an empty right hemiscrotum. On examination, the left testis had normally descended into the scrotum, but the right hemiscrotum was empty. There was suprapubic mobile swelling measuring 2 cm by 2.5 cm (Figure 1). It was mobile from side to side but could not be guided into the right hemiscrotum. Its consistency was akin to that of the left scrotal testis. The overlying skin was normal. The hernial orifices and the phallus were normal. An ultrasound confirmed it was similar to the left testis. Diagnosed as ectopic testis, the right inguinal canal was explored through a skin-crease incision extending to the

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case report
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Figure 1. Testis in the penopubic region.
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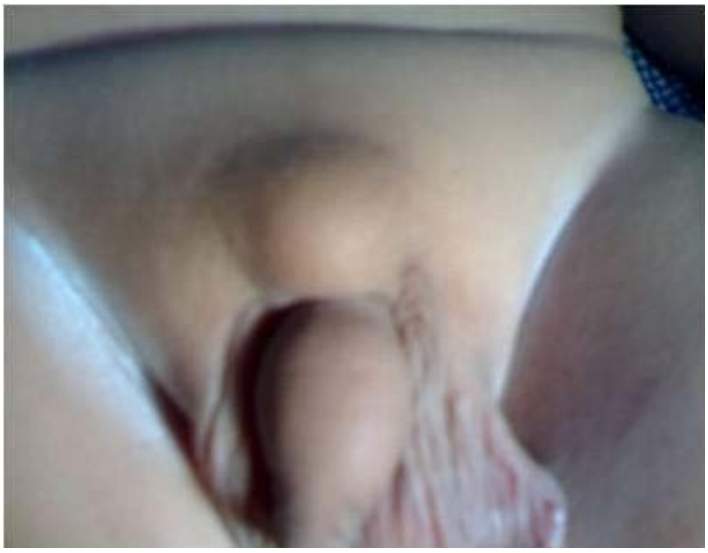

Figure 2. Testis before fixing in the subdartos pouch.
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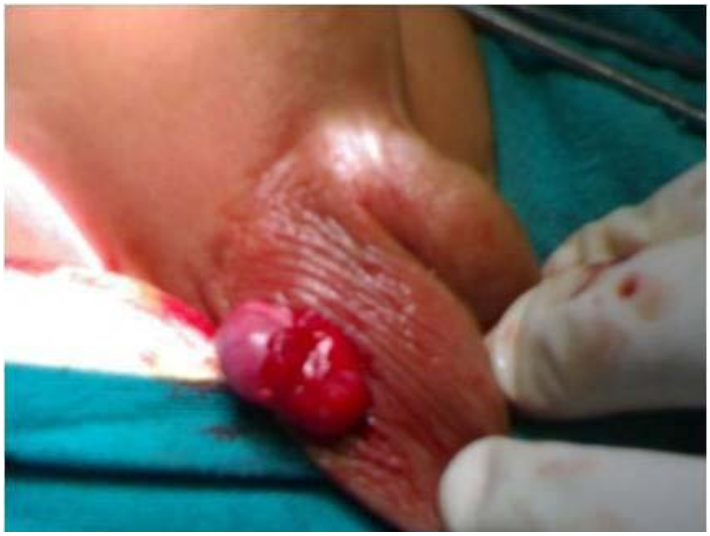

pubic tubercle. The spermatic cord, after emerging from the superficial inguinal ring, was going medially upwards. It was lying underneath the fascia of Scarpa. There was no hernial sac; however, a remnant of processus vaginalis was found, which was taken down. There was no gubernacular structure attached to the testis. There were minor fibrous adhesions around the testis. The testis and spermatic cord were easily mobilized and fixed in the scrotum in a subdartos pouch, without tension (Figure 2). The postoperative period was uneventful. At a 6-month follow-up, the testis was in the scrotum and of normal size.

DISCUSSION

Of the 5 sites described for ectopic testis (i.e., superficial inguinal pouch, femoral, perineal, transverse [contralateral hemiscrotum], and penopubic), the penopubic testis is the rarest, with an incidence of 0.6% [2]. In 1959, Collins [3] had recorded 9 cases of penopubic/penile testes, including 2 of his own. Since then, anecdotal cases of penopubic/penile testis have been reported [2,4-9]. Normal testicular descent occurs sequentially in 2 phases: transabdominal and inguinoscrotal [10]. Each phase has a different anatomical mechanism and is under the influence of different hormones. A key structure in the transabdominal phase is the gubernaculum (genitoinguinal ligament). The transabdominal phase of testicular descent is regulated anatomically by the vector sum of traction by the cranial suspensory ligament and the gubernaculum. The inguinal abdominal wall muscles form around the mesenchymal end of the gubernaculum, setting the stage for the future inguinal canal. Regression of the cranial suspensory ligament and gubernacular swelling reaction (enlargement of the caudal end of the gubernaculum by mitosis and deposition of hyaluronic acid) is under the influence of androgens and Müllerian inhibiting substance (MIS). The swelling reaction of the gubernaculum holds the testis very close to the future internal inguinal ring.

For the inguinoscrotal phase of testicular descent, Hutson proposed that the genitofemoral nerve acts as a second messenger for androgen by releasing a calcitonin gene-related peptide (CGRP), which provides the chemotactic signal for the gubernacular migration to the scrotum. Gubernaculum acts like a radar, taking the testis to the scrotum. Some very rare sites of ectopic testis have also been recorded. Gaur et al. [11] reported a case of subumblical testis. In his case, the testis had exited the internal ring and passed beneath the conjoint tendon to lie over the posterior rectus sheath above the linea semilunaris. Importantly, there was no gubernaculums found with the testis in this report. Redman et al. [12] reported a testis between the external and internal oblique muscle, superolateral to the internal ring. Similar cases were reported by E. Günel [13] and Mares [14]. Mares attributed this phenomenon to an absent

external inguinal ring in all his cases. Rao et al. [15] reported an ectopic testis in the iliac fossa, midway between the anterior superior iliac spine and the umbilicus, associated with arthrogryposis multiplex congenita. The testis lay between the external oblique aponeurosis and the fascia of Scarpa. Ameh et al. [16] described a neonate with scrotoschisis and bilateral extracorporeal testicular ectopia. Furtado et al. [17] and Murphy et al. [18] reported 1 case each of ectopic testis in the preperitoneal space near the umbilicus.

Various theories have been put forward to explain ectopic testicular migration. Lockwood [19] proposed his famous multiple insertion theory, describing the gubernaculum as a fibromuscular structure with insertions into the scrotum, perineum, Scarpa triangle, and the root of the penis. The scrotal insertion was considered dominant in normal descent, and an ectopic testis occurred when a dominant pull was directed by minor gubernacular tails. Sonneland [20], however, did not agree with Lockwood's theory and proposed that testicular ectopia occurs due to a break in the gubernaculum, leaving the testis radar a little less after its exit from the superficial ring. Backhouse [21] attributed ectopia to an ingrowth of fibrous tissue in the gubernacular mesenchyme, thereby hindering its attachment with the processus vaginalis. Hutson [10] proposed that the mislocation of the ipsilateral genitofemoral nerve causes the gubernaculum to migrate to the wrong site.

While the present theories for genesis of ectopic testis take into account the deviation of testis outside the external ring, the ectopic migration of testis before it crosses the external ring is not fully explained. These theories of descent do not include adequate data, either observational or experimental, to explain testicular ectopia, and presently, the mechanism of pathologic descent is largely unknown [22].

Although Lockwood's theory is often quoted in literature, our case does not support this view. There was no gubernaculum attached to the testis in our case. Sonneland's view is applicable to our case. The testis became rudderless after its exit from the superficial ring, as it lost its contact with the gubernaculum. It then occupied a position wherever the pressure in the fascial planes allowed. Once it stayed there for some time, local adhesions developed, keeping it in that location.

Whatever may be the mechanism of ectopic testis, a few clinical points are worth stressing again:

1. both the testis and the scrotum on the affected side in a case of ectopic testis are usually well developed, and in the case of undescended testis, the ipsilateral scrotum is often poorly developed [4],
2. the ectopic testis does not descend by itself; hence early surgery is advocated [15],
3. at surgery, it is mandatory to follow the spermatic vessels down to their ends [11], and
4. the long spermatic cord makes orchidopexy easy [13,15].

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