

The Prevalence of Bell Clapper Anomaly in the Solitary Testis in Cases of Prior Perinatal Torsion

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Purpose: Bell clapper anomaly is associated with an increased risk of intravaginal testicular torsion. However, perinatal torsion is thought to be secondary to an extravaginal process. We quantified the contralateral prevalence of bell clapper anomaly in children found to have atrophic testicular nubbins secondary to presumed torsion during gestation to better define the subsequent risk of metachronous testicular torsion.

Materials and Methods: Inspection results for the presence of contralateral bell clapper anomaly was recorded by a single surgeon in 50 consecutive cases in which exploration for nonpalpable testes revealed a testicular nubbin. For comparison data were collected in 27 consecutive cases of acute testicular torsion. Anatomy of the normal contralateral testis was compared between the 2 groups.

Results: Average age at surgery in the perinatal torsion group was 15 months vs 12.7 years in the acute torsion group. One case of partial contralateral bell clapper anomaly was discovered in the perinatal torsion group but no complete anomaly was found. In contrast, in older boys with acute testicular torsion complete bell clapper anomaly was found in 21 of the 27 contralateral testes (78%).

Conclusions: In older boys with acute testicular torsion contralateral bell clapper anomaly is highly prevalent, supporting the standard practice of contralateral testicular fixation in this clinical situation. However, the prevalence of contralateral bell clapper anomaly is exceedingly small in cases of monorchism after perinatal torsion, substantiating an insufficient risk of subsequent torsion to justify routine fixation of the solitary testis.

Key Words: testis, spermatic cord torsion, cryptorchidism, orchiopexy, age of onset

MONORCHISM is typically the result of testicular atrophy/regression from a presumed perinatal ischemic event or torsion.¹ These patients are initially diagnosed with unilateral nonpalpable cryptorchidism. However, in some patients a testicular nubbin can be palpated during careful physical examination using anesthesia or in a cooperative patient

while awake, and contralateral testicular hypertrophy is usually present.¹ Surgical exploration of these cases reveals hypoplastic wolffian structures in 90%, supporting the concept of testicular regression.²

Whether perinatal torsion is an intrauterine/prenatal or a neonatal event, it is widely agreed to be an extravaginal event except in a few

Abbreviations and Acronyms

TV = tunica vaginalis

TVPL = TV parietal lamina

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reports of acute intravaginal perinatal torsion.^{3,4} In contrast, acute torsion later in life is almost always intravaginal. While there is a general consensus to perform contralateral prophylactic orchiopexy for acute testicular torsion regardless of patient age at presentation, evidence for this practice is not as robust in the setting of exploration for unilateral monorchism. Central to this debate is the proper risk assessment of subsequent torsion in the solitary testis, which is almost exclusively an intravaginal process in the older male secondary to bell clapper anomaly or less commonly a generous mesorchium.⁵

To assess this risk of subsequent torsion we present what is to our knowledge the largest study to date assessing the prevalence of bell clapper anomaly in cases of solitary testis secondary to presumed contralateral perinatal torsion.

METHODS

From 2006 to 2012 the presence or absence of bell clapper anomaly in 50 consecutive boys was systematically recorded in the case log of a single surgeon (HGR). The patients were undergoing exploration for a nonpalpable testis with contralateral prophylactic orchiopexy of a solitary testis after simultaneous discovery of unilateral testicular atrophy. In the same fashion this anatomical information was also recorded from 2008 to 2013 in 27 consecutive cases of acute testicular torsion. With institutional review board approval we reviewed these case logs to determine the prevalence of bell clapper anomaly in each cohort as a predictor of the future risk of intravaginal torsion.

Bell clapper anomaly was confirmed when the TV completely encircled the testis, epididymis and distal spermatic cord rather than attaching to the posterolateral aspect of the testis and base of the epididymis. Thus, the testis and epididymis hung freely in a sac created by the TV (fig. 1).

Basic demographic variables were extracted from the medical record along with a review of final pathological findings and operative reports. We used basic descriptive statistical techniques to analyze the data.

RESULTS

The table lists basic demographics and laterality. There was a left predominance in the acute torsion cohort with no significant laterality predominance in the monorchism group. As expected, patients with acute torsion were much older than those with monorchism (mean age 12.7 vs 1.3 years). Black patients predominated in the acute torsion group at 52% while white patients accounted for 58% of those with monorchism. However, the numbers were too small to determine any meaningful racial associations. The racial distribution of acute cases could theoretically reflect the demographic distribution near our institution and family prioritization of acuity and proximity.

In the 50 cases of monorchism ipsilateral nubbins identified on scrotal exploration were removed for pathological evaluation. In 76% of these cases grossly evident hemosiderin deposits were identified with the testicular remnants at scrotal exploration, making laparoscopy unnecessary. In 24% of cases without definitive hemosiderin deposits laparoscopy was performed, which confirmed the absence of a viable undescended intra-abdominal testicle. In all cases specimens removed at scrotal or inguinal exploration showed histological evidence of a torsed testicle remnant, including atrophic wolffian structures, spermatic cord, fibrotic nodule, calcification or hemosiderin deposits, on final pathological assessment. No contralateral monorchid testes for which prophylactic orchiopexy was done showed true, complete bell clapper anomaly. One testicle demonstrated a partial anomaly.

All torsion was intravaginal in the 27 cases of acute testicular torsion and associated with ipsilateral bell clapper anomaly. The contralateral testicle also showed a complete anomaly in 21 of 27 cases (78%) (fig. 2). There was no case of acute perinatal torsion in this cohort.

DISCUSSION

Testicular torsion, more accurately described in the early literature as torsion of the spermatic cord, was first recorded by Nicoladoni in 1885 in an adolescent with an undescended testicle.⁶ Many theories of etiological predisposition were proposed with the term floating testicle the most common descriptor.⁷ It was not until 1957 that the term bell clapper became commonplace in the English literature but its origin is unclear.⁸ Regardless of etymology, the definition and proper recognition of bell clapper anomaly is critical to determine the risk of metachronous torsion beyond the neonatal period of about 1 month of life.

The definition of bell clapper anomaly depends on the posterior attachment of the TVPL or, more exactly, the posterior reflection point of the parietal and visceral layers of the TV. A normal testicle has posterolateral attachment of the TVPL to the epididymis and posterolateral aspect of the lower pole of the testis. In contrast, in patients with the bell clapper anomaly the attachment of the TVPL completely encircles and attaches more proximal to the distal spermatic cord. Thus, the testis, epididymis and distal spermatic cord hang freely in the TV up to the point of the TV visceral layer reflection (fig. 1). This leaves the testicle at risk for intravaginal torsion secondary to twisting of the distal unattached spermatic cord. Intermediate variations of this condition were reported less

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