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Controversies in the management of neonatal testicular torsion: A meta-analysis☆,☆☆

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ABSTRACT

Objective: This meta-analysis seeks to discern the optimal management strategy in neonatal testicular torsion (NTT).
Methods: Reviewed all English-language articles published between 2005 and 2015 in Medline/Pubmed that had a defined diagnosis of NTT within the first thirty days of life, and discussed specific surgical and nonsurgical management. Exclusion criteria were non-English literature, case reports, case studies, and failure to clearly describe the management of NTT. Data from 9 studies were analyzed, individually and together as pooled data, using a random effect model with a random intercept to estimate the pooled proportions of interest. Results are presented with 95% confidence interval. All analyses were done in SAS 9.4®.
Results: 9 publications met criteria for this analysis with a total of 196 patients. Bilateral testicular torsions (n = 14) were less common as compared to right/left testicular torsion (n = 85/97). Asynchronous NTTs (n = 9) were more common than synchronous NTTs (n = 2). There was a higher incidence of NTT in neonates delivered by vaginal delivery (n = 110) as compared to those delivered by c-section (n = 25). Extravaginal torsion (n = 54) is far more common than intravaginal torsion (n = 2). Full-term neonates (n = 122) have a higher incidence of NTT as compared to preterm neonates (n = 9). A total of 15 testicles were salvaged. Of the salvaged testicles 2 were documented as prenatal, 10 postnatal and 3 were undocumented. A strategy of bilateral exploration allows for salvage of about 7% of ipsilateral testicles and prevent asynchronous torsion in about 4% of neonates.
Conclusions: Based on our population, between 8–12% of patients would benefit from bilateral exploration at the time of diagnosis. We recommend urgent bilateral exploration with orchiopexy of the contralateral testicle in order to avert anorchia.
Type of study: Systematic review.
Level of evidence: Level 5 meta-synthesis (Evidence from systematic reviews of qualitative and descriptive studies).

Contents

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Testicular torsion management has plagued urologists and surgeons alike. With the incidence of neonatal testicular torsion (NTT) at 6.1 per 100,000 live births, prospective studies of management strategies are difficult to obtain [1–3]. Furthermore, the presumed need of prompt diagnosis has not allowed for proper prospective studies to identify the correct course of action when faced with a neonate with testicular torsion. NTT is defined as a torsion that occurs within the first thirty days of life which can occur in the prenatal period, or within 30 days of postnatal life. Torsion can be further broken down to extravaginal and intravaginal. Extravaginal, which is most commonly seen in neonates, is when the testis, epididymis, and tunica vaginalis twist on the spermatic cord [4–6]. In contrast, intravaginal torsion occurs secondary to a bell clapper deformity where the tunica vaginal inserts onto the spermatic cord higher than normal allowing the testes to rotate on itself [4–6]. Even with an early diagnosis, the salvage rate for torsions has been reported at about 5% [7]. As a result of this poor salvage rate, the necessity of urgent surgical intervention has been heavily debated. Owing to the perceived unlikely salvage, many surgeons have adopted a wait and watch approach. Others, however, feel that immediate exploration and orchiopexy of the contralateral side simultaneously or after a few months are prudent. This meta-analysis seeks to discern the optimal management strategy in NTT based on a review of recent results in the management of this condition.

1. Materials and methods

We reviewed all English-language articles published between 2005 and 2015 in Medline/Pubmed and SCOPUS that had a defined diagnosis of NTT within the first thirty days of life, and discussed specific surgical and nonsurgical management. Key words used for the search were: neonatal testicular torsion, neonatal testicles, neonatal orchiopexy, and perinatal torsion. Exclusion criteria were non-English literature, case reports, case studies, unable to clearly define neonatal testicular torsion within the first thirty days and failure to clearly describe the management of NTT (Fig. 1).

The main characteristics of the studies evaluated were the mode of delivery, laterality, gestational age, and management of ipsilateral and contralateral testes. Treatment strategies were classified into 4 groups to facilitate comparison (Fig. 2). Group 1: Bilateral exploration; Group 2: Exploration of affected side only; Group 3: Exploration of contralateral side only; and Group 4: Observation only. Other aspects reviewed were prenatal torsion, postnatal torsion, intravaginal torsion and extravaginal torsion. Data from selected studies were analyzed using a random effect model with a random intercept to estimate the pooled proportions of interest. Results are presented with 95% confidence interval. Each study was evaluated separately as well as pooled together to form combined data. All studies included in the meta-analysis were

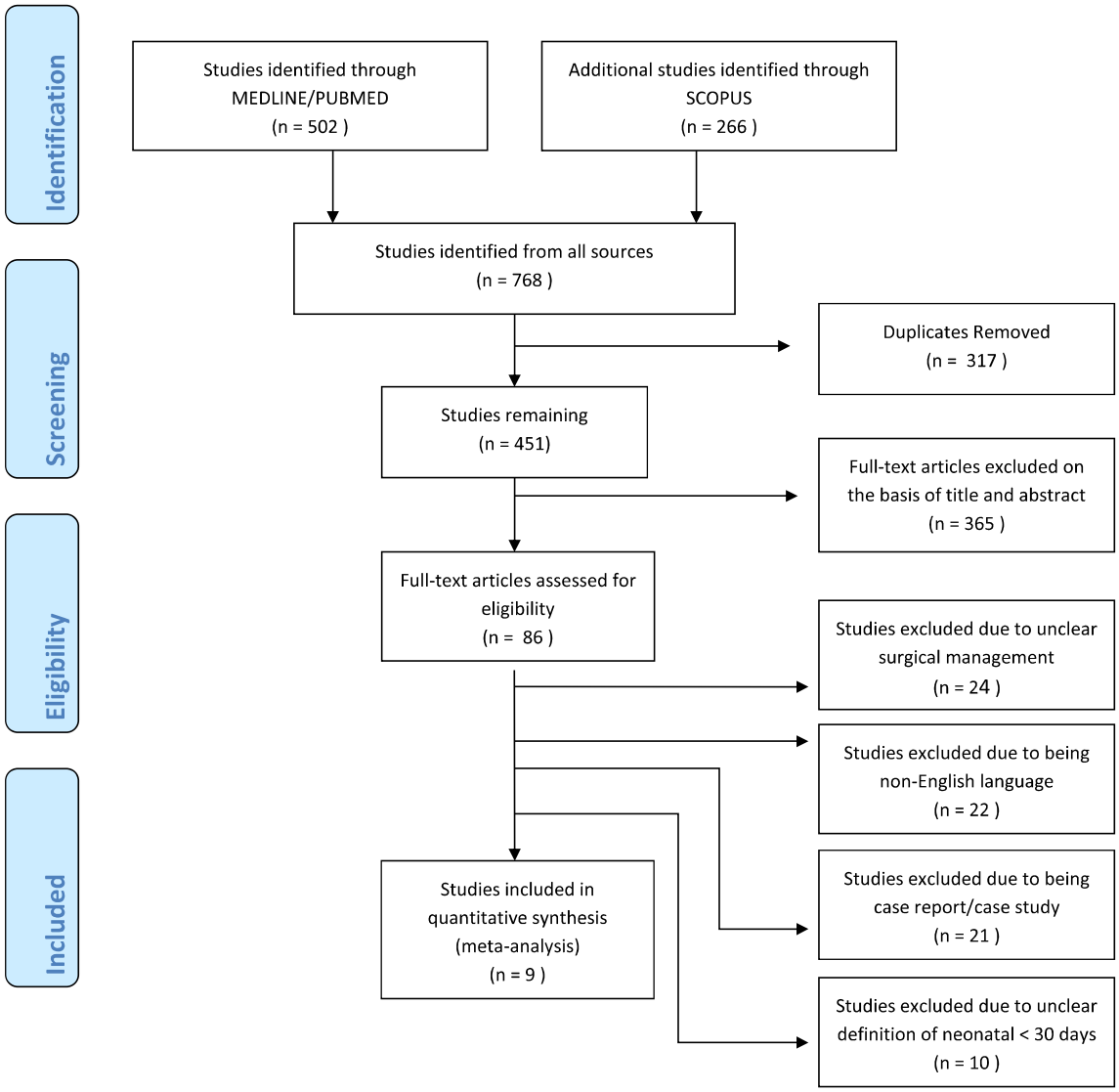

Fig. 1. Study selection flowchart.

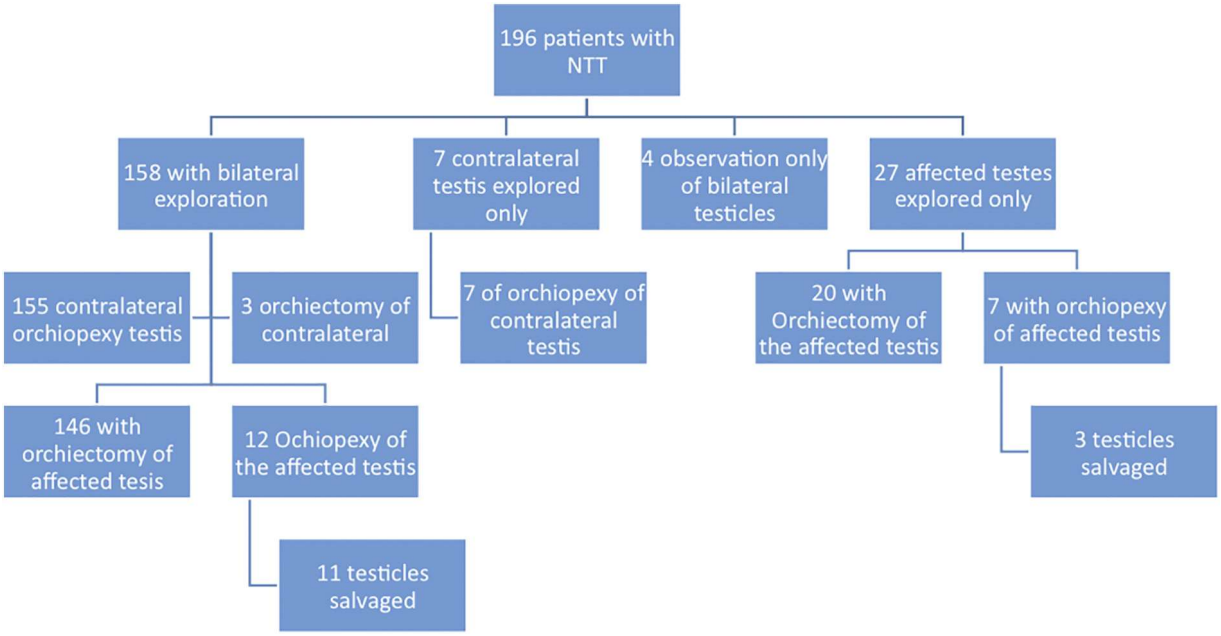

Fig. 2. Flowchart of NTT systematic review treatment strategies and outcomes.

used to estimate pooled confidence intervals (CI). The 95% CI discussed throughout the results represents the upper and lower limit of the estimated proportion. That is we are 95% confident that the proportion of patients who had the result/procedure is between the upper and lower limits set by the CI. The wider the CI the lower is the possibility of a Type 1 error; however, narrower CIs have a better accuracy. No p-value calculations were possible. All analyses were done in SAS 9.4®.

2. Results

9 publications met criteria for this analysis with a total of 196 patients (Table 1). Results are summarized in Tables 2 and 3. Of these 196 patients, there were torsion of the right testicle in 85 patients [pooled proportion 0.43; Confidence Interval (CI) (0.34–0.53)], 97 patients on the left [pooled proportion of 0.49; CI of (0.39–0.59)] and 14 bilaterally [pooled proportion of 0.7 and CI (0.03–0.15)]. A total of 15 testicles were salvaged. Of the salvaged testicles 2 were documented as prenatal, 10 postnatal and 3 were undocumented.

Of the 196 patients 110 of these infants were born via vaginal delivery [pooled proportion 0.54; CI (0.17–0.87)], 25 via c-section [pooled proportion 0.12; CI (0.06–0.23)], and 61 unknown. There were 122 term infants; defined as greater than 37 weeks [pooled proportion 0.70; CI (0.07–0.99)] vs 9 preterm infants; less than 37 weeks [pooled proportion 0.02; CI (0.01–0.10)] and 65 unknown gestational age.

98 patients had a documented prenatal torsion [pooled proportion 0.56; CI of (0.13–0.92]; 35 patients had a documented postnatal torsion [pooled proportion 0.10; CI (0.03–0.31). There were 63 undocumented.

2 patients had documented intravaginal torsion [pooled proportion 0.01; CI (0.00–0.05)] and 54 patients had extravaginal torsion [pooled proportion 0.28; CI (0.03–0.85)]. 140 of the 196 had an undocumented type.

158 patients had bilateral testes explored (Group 1) while 27 only had the affected testicle explored (Group 2), 7 patients had only contralateral testicle explored (Group 3) and 4 were observed without surgical management (Group 4). In Group 1, 146 of these patients had an ipsilateral (I/L) orchiectomy and bilateral testes explored with a pooled proportion of 0.75 and confidence interval of (0.49–0.9). 12 patients had orchiopexy of the affected testicle with an exploration of the contralateral testicle with a pooled proportion of 0.06 and CI of (0.02–0.18). 11 of the patients who had ipsilateral orchiopexy and exploration of the contralateral testicle had the testis salvaged with a proportion of 0.06 and CI of (0.02–0.14). In Group 2, 20 patients had orchiectomy of the affected testicle only, with a pooled proportion of 0.1 and CI (0.07–0.15). 7 patients underwent orchiopexy of the affected teste with a pooled proportion of 0.03 and CI of 0.01–0.11. 3 of these testicles were salvaged in the group with ipsilateral explored only with a CI of 0.01 (0–0.6). 4 patients (Group 4) were observed without surgical management.

Table 1
Article demographics.

Author	Yr. Published	Journal	# of patients in study
Abraham et al.	2015	Journal of Maternal–Fetal and Neonatal Medicine	28
Al-Salem, Ahmed	2007	Journal of Pediatric Surgery	11
Arena et al.	2006	International Journal of Urology	7
Djahangirian et al.	2010	Journal of Pediatric Surgery	44
Jensen et al.	2015	Danish Medical Journal	13
John et al.	2008	Acta Paediatrica	24
Kaefer et al.	2015	Journal of Pediatric Urology	37
Kaye et al.	2007	Journal of Urology	15
Mano et al.	2013	Urology	17

Table 2
Data results.

	No. patients documented	Pooled Proportion	Pooled 95% CI
Left Testicular Torsion	97	0.49	0.39–0.59
Right Testicular Torsion	85	0.43	0.34–0.53
Bilateral Testicular Torsion	14	0.07	0.03–0.15
Asynchronous	9	0.04	0.00–0.05
Synchronous	2	0.01	0.01–0.11
Vaginal delivery	110	0.54	0.17–0.87
C-section	25	0.12	0.06–0.23
Full term	122	0.70	0.07–0.99
Preterm	9	0.02	0.01–0.10
Extravaginal	46	0.28	0.03–0.85
Intravaginal	2	0.01	0.00–0.05

Table 3
Surgical management characteristics.

	B/L Testes Explored	I/L Testis Explored Only	C/L Testis Explored Only	Observed
Number of patients	158	27	7	4
Pooled Proportion /(CI) of ipsilateral ORCHIECTOMY	0.75 (0.49–0.9)	0.1 (0.7–0.15)		
	N = 146	N = 20		
[proportion of documented]	[92.4%]	[74%]		
Pooled proportion/ (CI) of ipsilateral ORCHIOPEXY	0.06 (0.02–0.18)	0.03 (0.01–0.11)		
	N = 12	N = 7		
[proportion of documented]	[7.6%]	[35%]		
Pooled proportion/ (CI) of SALVAGED Testicles	0.06 (0.02–0.14)	0.01 (0–0.06)		N = 1
	N = 11	N = 3		
[proportion of documented]	[91.2%]	[42.9%]		

Of the 15 testicles that were salvaged 2 of those were in a documented prenatal torsion with a pooled proportion of 0.01 (0.00–0.07) and 10 were documented postnatal torsion CI (0.05–0.12). 3 were unknown.

3. Discussion

Owing to neonatal testicular torsions rarity, the proper management has been difficult to ascertain. Currently, the most common practice currently is to perform bilateral exploration with orchiopexy of the unaffected testicle to prevent the devastating effects of anorchia, should asynchronous torsion occur. Based on this meta-analysis it appears that bilateral exploration (Group 1), by allowing effective comparison of the affected testicle with the opposite normal side leads to lower orchiopexy rates (12/158) but higher ipsilateral salvage rates (11/12). In Group 2, where only the affected testicle is explored, there appears to be a tendency to perform a higher rate of orchiopexy (7/27) with lower salvage rates (3/7). Based on these findings we recommend a bilateral exploration in NTT. Our study found 7%; 14 of 196 patients, had bilateral torsions with 9 or 4% reported as asynchronous. This is an interesting finding based on the fact that other studies have found synchronous torsion to be much more common [4]. Abraham et al. had similar findings in their single-center experience with all of their 5 bilateral torsions being asynchronous events [8]. This finding may push towards orchiopexy of the unaffected testicle in order to prevent a catastrophic event of anorchia. Yet, as described in various other studies the risks of urgent surgical intervention in the neonate, especially anesthetic risk, must be heavily weighed prior to proceeding with surgery [8–10]. With a Group 1 strategy of bilateral exploration, there would be a 7% salvage of the ipsilateral testicle and insurance against the 4% chance of asynchronous torsion. Overall this strategy would benefit between 7% and 12% (15 ipsilateral torsion salvage and 9 asynchronous torsion prevention of the total of 196) of neonates prone to developing NTT.

There have been multiple theories regarding why torsion would occur in the first place. It appears that intrauterine stress may be a significant risk manifested by prolonged or difficult labor, high birth weight, breech presentation, preeclampsia, or vaginal delivery to name a few described in previous studies [5,6,10]. Similar to other studies we found vaginal delivery to be associated NTT. It has been postulated that hypermobility of the tunica vaginalis within the scrotal sac when exposed to an extreme cremasteric reflex during delivery or in utero, may induce a torsion [5,6,10,12]. This may also explain why the meta-analysis population, which is consistent with other reports, presented with extravaginal torsions as opposed to intravaginal. Based on our data NTTs occurred most commonly in those born vaginally. This could also be explained by the fact that most of our study population were born vaginally as opposed to c-section. Factors around these deliveries such as prolonged rupture of membranes or other health risks were not adequately described to comment and therefore no firm stance can be provided on their role in NTT. Further study should be done on high-risk pregnancies and intrauterine stress as a possible contributing factor for NTT.

In our population, we found that no statistical difference in the occurrence of torsion in the left as opposed to the right. This is in-line with more recent studies in which there was no difference in torsion occurring on one side greater than the other [5,11]. It had been previously reported that NTT occurred more frequently on the left possibly owing to differences in the testicular vasculature [5]. This statement was not confirmed via this meta-analysis.

In contrast to many other studies two of the salvaged testes were documented as prenatal leading us to believe that increased surveillance of testicles at time of delivery coupled with early surgical intervention may actually increase the salvagibility [10]. This is, however, a hypothesis not based on literature evidence.

It has been postulated that removal of the affected testicle may not be best practice as there is some evidence that the testicle may maintain endocrine function even when the spermatogenic function has been adversely affected [4,11]. Yet, other studies have suggested that leaving the torsed testicle may be a nidus for infection, be a source of malignancy later in life, or become a source for testicular tissue antibodies; therefore, it is often recommended that the nonviable torsed testicle should be removed [4,13,14]. This aspect should be studied further to assess the true risk versus benefit. In our study, all of the testicles that were observed underwent atrophy suggesting that there is no significant benefit to surgically explore clearly, nonviable torsed testicles as the risk for formation of autoantibodies may be an overestimate.

Our study has its limitations. We relied on others' documentation as well as lack thereof for our data which also may be biased in nature. Also, the specific characteristics of torsed testicles that underwent an orchiopexy were not clearly defined which affects the ability to determine why certain testicles were salvageable and others were not. The timing of surgical intervention was lacking further affecting the ability to establish salvage rates based on the diagnosis of NTT to OR time. Owing to the rarity of neonatal testicular torsion prospective studies would be difficult to manage.

4. Conclusions

Any suggestion of viability urgent exploration should be considered not only in the attempt to salvage the affected testicle but to prevent the devastating effects of a bilateral torsion with subsequent anorchia. It appears that exploration of the contralateral testicle is the current standard of care. Based on our population, 8–12% of patients would benefit from bilateral exploration at time of diagnosis. We recommend urgent bilateral exploration with orchiopexy of the contralateral testicle in order to avert anorchia.

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