Title

Management Strategies for Bilateral Intra-abdominal Testes: Insights from a Tertiary Paediatric Centre

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Abstract

Introduction

Cryptorchidism, occurring in 2-4% of male newborns, necessitates orchidopexy to mitigate associated risks, preferably between ages 3-12 months. While unilateral cases are common, bilateral intra-abdominal testes (BIAT) present unique challenges. This study reviews outcomes following bilateral Fowler-Stephens orchidopexy (FSO) at a paediatric surgical tertiary centre, aiming to enhance preoperative counselling and contribute to management consensus.

Methods

A retrospective analysis was conducted at a tertiary paediatric centre, identifying BIAT cases between 2005 and 2021. Clinical records of patients undergoing bilateral staged orchidopexy were reviewed. Primary endpoints included testicular size and position, with secondary outcomes examining staged versus non-staged procedures.

Results

We identified thirty patients who underwent Fowler-Stephens orchidopexy (FSO) for bilateral intra-abdominal testes (BIATs) between 2005 and 2021. The procedures were performed using

both open surgery (n=6) and laparoscopic techniques (n=24), based on consultant preference. Among these patients, three underwent single-stage procedures, with 2 out of 3 having both sides operated on the same day. For the remaining twenty-seven patients, staged procedures were performed. In 22 out of 27 cases, the first stages were conducted bilaterally during the same operation. The second stages were performed synchronously in 3 out of 27 patients and on a metachronous basis in 24 out of 27 patients. The mean age at the time of the first procedure was 1.7 years, ranging from 3 months to 10 years. Outcomes revealed a success rate of 82%, with 14% atrophy and 3.5% ascent rates. Comorbidities were noted in 50% of patients, and only eight boys underwent endocrine evaluation.

Conclusion

Management of BIATs requires tailored approaches, with staged FSO offering favourable outcomes. Further research is needed to establish consensus guidelines for optimal management strategies.

Keywords

Undescended testes, Fowler-Stephens, Orchidopexy

Abbreviations

BIAT – Bilateral Intraabdominal Testes

1.Introduction

Cryptorchidism affects 2-4% of male newborns (1,2), however 80% of these testes descend within the first 3 months after birth reducing the true incidence of testes requiring an orchidopexy to 1% (3). The Royal College of Surgeons commissioning guide recommends performing orchidopexy for palpable undescended testes between the ages of 6 to 18 months, if feasible (4). In boys with impalpable testes, the testis may be absent, ectopic, located in the inguinal canal, or within the abdominal cavity, the latter is termed 'intra-abdominal testes' (IAT) (5-7). Fowler-Stephens orchidopexy (FSO) is a widely used surgical approach to mobilize IATs into the scrotum, often performed as a two-stage procedure. The reported success rate of FSO ranges from 65-85%, with failure defined as testicular atrophy or ascent (5,6,8-13).

Approximately 10% patients present with bilateral intra-abdominal testes (BIAT) (14). While the technical aspects of management do not differ in these cases, additional considerations include the timing of staged surgeries on each side, the risk of anorchia in the event of bilateral atrophy, and the potential implications for puberty and fertility in affected individuals. There is no published consensus on the optimal management pathway for boys with BIAT, and the

existing literature is challenging to analyse due to the lack of differentiation between unilateral and bilateral IAT cases. **Specifically, there is a dearth of data on the possibility of anorchia resulting from bilateral atrophy, as well as possible risk reduction strategies to mitigate this complication and allow pubertal progression.**

The aim of this study was to report the outcomes of patients undergoing bilateral FSO at a tertiary paediatric surgical centre with a view to identifying practical insights that can enhance preoperative counselling for patients. A secondary aim was to contribute to the accumulation of data necessary for establishing a consensus on the optimal management of BIAT.

2.Methods

Patients with BIAT treated at Alder Hey Children's Hospital between January 1, 2005, and March 31, 2021 were identified using ICD10 codes Q53.1-9 for undescended testis. The medical records were reviewed: all patients whose records confirmed the diagnosis of BIAT, who underwent one or two stage bilateral Fowler- Stephens orchidopexy (OPSC9 N08.3-4) and had documented follow up were included in the study. **Patients who underwent any other form of orchidopexy were excluded.** The primary endpoint was subjective analysis of testicular size and position by a clinician during follow-up visits. **Secondary outcomes were categorized based on synchronous versus metachronous procedures**. Success was defined as the

presence of palpable testicular tissue in the scrotum before puberty, after a minimum follow-up period of three months. **Patients with Prune Belly syndrome were excluded as management of their BIATS was influenced by more clinically urgent comorbidities in most cases**. A summary of their treatment and outcomes has been published separately (15).

3. Results

3.1 Surgical Strategy

Thirty patients who underwent Fowler-Stephens orchidopexy (FSO) for bilateral intra-abdominal testes (BIAT) between 2005 and 2021 were identified. Care was provided by nine consultants; perioperative protocols were not standardized. The procedures were performed via an open (n=6) or laparoscopic approach (n=24), based on consultant preference. **Three underwent single-stage procedures because the testis achieved enough length after dissection to reach a good scrotal position**, 2 of whom had both sides operated on at the same sitting. For the remaining 27 patients, staged procedures were performed. **In these staged procedures, the testicular vessels were clipped or ligated in the first stage and the testis was brought down to the scrotum in the second stage**. In 22 out of 27 cases, first stage FSO was conducted bilaterally during the same operation. The second stage FSO were performed synchronously in 3 patients and metachronously in 24 patients (Figure 1). In 4 out of 24 patients, the second-stage procedure on the second side was postponed until after puberty for various reasons (see section 3.2). The mean age at the

time of the first procedure was 1.7 years (range 3 months to 10 years), late referral delayed surgery in six cases.

3.2 Testicular outcome

Testicular outcomes were assessed during follow-up appointments at outpatient clinics (Figure 2). Twenty-two (73%) boys successfully completed treatment with two normal-sized testes in their scrotum. However, one of the 22 boys required a unilateral redo orchidopexy due to testicular ascent. Another boy experienced ascent of one testis to the superficial inguinal ring, which was considered unsuitable for a redo procedure. Currently, this patient awaits puberty, at which point revision or orchidectomy will be considered. Four patients who completed treatment before reaching puberty had unilateral (n=3) or bilateral (n=1) testicular atrophy**. Hormonal support through puberty was required for the patient with bilateral atrophy.** In four patients who had unsuccessful second stage FSO surgery on the first side, a decision was made to leave the remaining testis within the abdomen to guide them through puberty, to avoiding pre-pubertal anorchia. The reasons for surgical failure included testicular atrophy (n=3) and orchidectomy for a dysplastic testis (n=1). Of these patients, only one has undergone completion of the second side at the time of writing, which was successful. (Figure 2). Table 1 shows the outcomes in relation to the type of operation performed.

To date, 57 out of 60 testes have undergone surgical treatment, with an atrophy rate of 14% (8/57) and an ascent rate of 3.5% (2/57), one of which was successfully revised. Additionally, one orchidectomy was performed for a dysplastic-looking testis. The median follow-up duration was 16 months, range: 3–54 months , after the final procedure.

3.3 Comorbidity

Within this cohort, 14/30 children had associated abnormalities, including 5 with endocrine or genital abnormalities **but no cases of disorder of sexual differentiation were identified (Table 2**). **These patients were referred to the endocrine team for hormonal assessment and continued follow up.**

3.4 Endocrine Outcome

Eight boys were referred for endocrine evaluation out of which 3 had associated genital abnormalities. Two post pubertal and 2 pre-pubertal boys with successful bilateral orchidopexies were found to have abnormal endocrine function. Conversely, one patient with a single testis had a normal endocrine profile.

4. Discussion

In this series, the largest in the literature, the outcomes for 30 patients undergoing management of bilateral intra-abdominal testes (BIAT) over a 16-year period are described. We report a success rate of 73% whereby success is defined as 2 palpable testes in the

scrotum before the child reaches puberty. Specifically, 8 (14%) testes atrophied leaving one boy anorchic. The findings highlight heterogeneity in the management of BIAT, likely reflecting variations in general practice.

The study identifies a higher atrophy rate than those reported in other studies examining BIAT outcomes as summarised in Table 3, although the atrophy rate is consistent with meta-analyses of Fowler-Stephens orchidopexy (FSO) overall (16). Hvistendahl and Algaratnam report series including both bilateral and unilateral cases (11,17). Hvistendahl *et al.* reported a 14% overall atrophy rate but did not specifically address bilateral cases. They suggested that bilateral cases had favourable outcomes compared to unilateral cases, possibly due to the lower mean age at surgery in bilateral cases, although statistical analysis was not conducted. Algaratnam *et al.* observed atrophy in 3.2% of bilateral cases versus 11.3% of unilateral cases, but this difference did not reach statistical significance, likely due to the small sample size. Furthermore, the median age did not differ between groups in Algaratnam's study, undermining Hvistendahl's suggestion of a lower atrophy rate in bilateral cases due to the younger age at surgery. None of the aforementioned studies included patients with unilateral atrophy who underwent delayed procedures until after puberty, nor did they mention this strategy. This series benefits from ample follow-up time (median 16 months, range: 3–54 months) using a pragmatic assessment of testicular outcomes in the absence of an objective

method due to its retrospective nature. This approach is consistent with many other series and may even overestimate atrophy rate, highlighted by the patient who had spontaneous puberty with what was assumed to be a solitary atrophic testis.

Assessing the impact of orchidopexy on an individual child's risk of testicular cancer presents challenges. Epidemiological studies investigating the link between undescended testes and malignancy are typically large but often lack detailed information on the preoperative position of the testis (18,19). BIAT could be associated with a higher risk of malignant transformation, although quantifying this relationship remains challenging due to the rarity of both BIAT and testicular tumours. Cortes *et al*. conducted a biopsy study on 1335 boys undergoing orchidopexy, finding a higher frequency of premalignant and malignant processes in BIAT and in children with abnormal external genitalia (20). As a result, paediatric surgeons are generally reluctant to leave a testis within the abdominal cavity if orchidopexy is not feasible, opting instead for orchidopexy or orchidectomy.

Moreover, children with BIAT are at risk of anorchia as a result of failed surgical management. With a reported atrophy rate of approximately 10-15%, bilateral failure of orchidopexy can lead to anorchia or very low testosterone levels, potentially inadequate for puberty (16). According to guidance from 2015, early endocrine assessment is recommended for all patients with bilateral

undescended testes, and this is now recommended practice in our centre (4).

Considering the risk of anorchia, we advocate for a synchronous first stage followed by a metachronous second-stage, as it allows for the option of delaying the second stage of the second testis until after puberty should the first side atrophy. This strategy was employed in four of our patients. Notably, one patient completed treatment after a delay of 6.1 years, which was ultimately successful. **It's important to note that if this strategy is employed, close follow-up until final orchidopexy is mandatory because of the risk of malignancy.** The remaining three patients await their delayed second-stage procedure. However, single-stage bilateral procedures may be suitable for children with significant comorbidities to reduce exposure to general anaesthesia. This approach was taken in a child with arthrogryposis, with a good outcome. Additionally, it is noteworthy that fertility in patients with BIAT is poor, even after successful orchidopexy, with azoospermia reported in over 50% of cases [20]. Thus, the aim of orchidopexies in these boys is to move the testes out of the abdomen and place them in the scrotum to allow self-examination, and to preserve their endocrine function.

Any consensus on the management of BIAT must consider all of the factors discussed above. Unfortunately, much of the literature on the outcomes of intra-abdominal testes does not clearly differentiate between unilateral and bilateral cases. A literature search revealed

only four studies specifically commenting on the outcomes of surgery for BIAT (11,17,21–22). Case series from Algaratnam, Safwat, and Hvistendahl distinguish between unilateral and bilateral IATs. Kaye describes a single-stage non-FSO technique for BIAT with success rates exceeding 90%. Where management of BIAT is described, there is typically a failure to address the specific challenges of managing these patients. Consequently, there is little guidance on management in the situation of testicular atrophy one side and incomplete staged orchidopexy on the other.

Several limitations of this review should be noted. Hospital coding may be unreliable, and so some cases with BIAT may not have been captured. Moreover, testicular outcomes were subjectively assessed by clinicians, and preoperative testicular size documentation was inconsistent. The small sample size precluded statistical analyses.

Conclusions

The management of bilateral intra-abdominal testes (BIAT) demands careful consideration, yet a published consensus remains elusive. Based on the findings of this study and existing literature, staged orchidopexy with metachronous second-stage procedures, performed at a young age, seems a relatively low-risk approach to BIAT management. Metachronous second-stage procedures should be spaced adequately to allow for assessment of the outcome of the first side before proceeding with the second procedure, as bilateral atrophy remains a possibility. **Moreover, Early endocrine**

assessment is recommended for all patients with BIATS regardless of the orchidopexy outcome.

Conflict of interest statement

None to declare

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Ethical Approval

None Required

Generative AI and AI-assisted technologies were NOT used in the preparation of this work

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Treatment Strategy	Total	Complete	Bilateral Good Outcome	Unilateral Atrophy	Bilateral Atrophy	Unilateral Ascent	Orchidectomy
Single Stage	3	3/3	3	0	0	0*	0
Staged Synchronous	2	2/2	1	1	0	0	0
Staged Metachronous	21	21/21	17	2	1	1	0
Delayed second side	4	1/4	0	3	0	0	1
Total	30	27/30	21	6	1	1	1

Table 1 – Patient outcome following Fowler Stephens orchidopexy for bilateral intra-abdominal testes according to treatment strategy.

* One patient in the single stage group required revision orchidopexy which was successful.

Embryological / Genetic	Genital/Endocrine
Trisomy 9	Micropenis
ASD, Pulmonary regurgitation	
Noonan's Syndrome	
11p13 deletion	Peno-scrotal hypospadias, Mullerian remnant
T21	
Coarctation	
Hydrocephalus	
Noonan's Syndrome	
Hepatic/Portal venous anomaly	Hypospadias, Partial androgen insensitivity
Beckwith Wiedemann syndrome, CHI, Cleft	
Palate	
ASD, VSD	
Malrotation, Pulmonary stenosis, ASD,	
Tracheomalacia	
ASD	Peno-scrotal Hypospadias
Tracheomalacia, Jejunal Atresia, Duplex	
Kidney, Arthrogryposis	
	Gonadotrophin deficiency
Club Feet	

Table 2: Associated abnormalities in the boys with BIAT

Study	Number of Patients		Outcome Bilateral (Unilateral)				Follow up (Weeks)	Timing of Stages (Bilateral Only)		
	Bilateral	Unilateral	Pending 2nd side	Atrophy	Ascent	%Success	Minimum (Median)	Single Stage	Synchronous	Metachronous
Algaratnam	19	75	-	1 (8)	5 (4)	80 (83)	12 (109)	0	11	8
Safwat	24	0	-	2	5	86	Not stated	6	Total of 18	
Hvistendahl	70 Total		-	-	-	82(80)	12 (12)		Not stated	
Kaye	21	0	-	2	0	91	26 (26)	19	1	1
This Study	30	0	3	8	1	73	14(101)	3	3	24
Total (Mean %)	94			12	11	82%		27	66	

385 **Table 3:** ***Summary of studies examining BIATs outcomes***

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Figure Legends

Figure 1: Cohort of BIAT

Figure 2: Outcomes of FS in BIAT.

*One case had successful orchidopexy on the other side.

Figure 1

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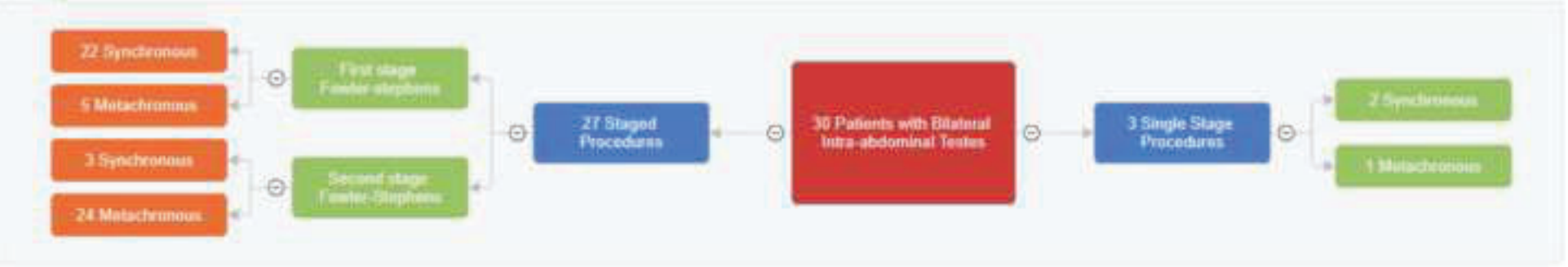

Figure 2