

Case Report

Preservation of testicular tissue after enucleation of pediatric mature teratoma: A case series of 7 testes in 6 children

Akihiro Kanematsu  and Shingo Yamamoto

Department of Urology, Hyogo College of Medicine, Hyogo, Japan

Abbreviations & Acronyms

β hCG = beta human chorionic gonadotropin
AFP = alpha fetoprotein

Correspondence: Akihiro Kanematsu, M.D, Ph.D., Department of Urology Hyogo College of Medicine, 1-1 Mukogawacho, Nishinomiya, Hyogo 663-8501, Japan. Email: aqui@hyo-med.ac.jp

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Introduction: A standard protocol for testis-sparing surgery for pediatric benign testicular tumors has not been established to date.

Case report: We treated 7 teratomas in 6 patients aged 12 years of younger. For 2 noncystic lesions, the spermatic cord was exposed via the transinguinal approach and clamped until the establishment of an intraoperative pathological diagnosis. The other 5 tumors in 4 infant patients were all preoperatively diagnosed with cystic teratoma, and we essentially exposed the testis directly via the scrotal approach and enucleated the tumor without clamping the cord. In every case, adequate amount of normal testicular tissue was preserved, without any local recurrence with 3-year minimum follow-up.

Conclusions: An unclamped enucleation of testicular tumor via the transscrotal approach, which is contraindicated in most testicular tumors, can be a treatment choice exclusively for preoperatively diagnosed mature cystic teratoma in infants.

Key words: children, enucleation, teratoma, testicular tumor.

Keynote message

An unclamped enucleation of testicular tumor via the transscrotal approach, which is contraindicated in most testicular tumors, can be a treatment choice exclusively for preoperatively diagnosed mature cystic teratoma in infants.

Introduction

Testis-sparing surgery has recently become the preferred treatment choice for pediatric benign tumors of the testis.^{1–6} However, because of the rarity of the disease, a standard protocol for surgical treatment has not been established to date. Moreover, the actual long-term preservation rate of the testicular tissue has not been adequately investigated. Herein, we present a single surgeon experience of tumor enucleation of 7 testicular mature teratomas in 6 patients.

Case report

The study was authorized by the ethics committee of the Hyogo College of Medicine. Between 2011 and 2016, we treated 7 teratomas in 6 patients aged 12 years old or younger. The summary of the cases is shown in Table 1. Two teratomas were in children aged over 10 years, in which malignancy was not completely excluded. The other patients were 4 infants and all were preoperatively diagnosed with cystic teratoma, including one bilateral case. Three infants initially presented a testicular cyst but a solid lesion appeared during follow-up, and all subsequently underwent surgery with the diagnosis of cystic teratoma.⁷ The initial tumor and cyst volume percentage to total ipsilateral testicular volume ranged from 16% to 86% (median 34, Table 1).

All tumors were enucleated. Intraoperative ultrasound was employed to evaluate the precise location of the tumor. After enucleation, the tunica albuginea was approximated by a 6-0 polyglactin suture after confirmation of benign teratoma by intraoperative pathological diagnosis.

Table 1 Clinical features of the cases

Case	Age (month)		At presentation	At surgery	Laterality	Serum marker		AFP	βhCG	Approach	Cord clamp	Pathology	Preoperative testis volume (mL)		Volume of tumor and cyst		Testis volume at final follow-up (mL)		Volume ratio of affected/nonaffected side	Follow-up (month)	Tanner stage at final follow-up
	At	At				Right	Left						Volume (mL)	Volume ratio†	Right	Left					
1	11 year				Left	1.6	<0.4			Inguinal	Yes	Mature teratoma	1	3.2††	1.73	0.54	8.9	5.7††	0.64	77	G5H5
2	12 year				Left	1.2	≤0.1			Inguinal	Yes	Mature teratoma	7.3	18.3††	6.9	0.37	10.3	7.7††	0.75	36	G5H5
3‡	1 year	1 year			Right	3.8	≤0.1			Inguinal	No	Mature cystic teratoma	1.3††	NA	0.23	0.18	0.43††	NA	NA	82	G1H1
4	1 month§	8 month			Left	3.8	≤0.1			Scrotal	No	Mature cystic teratoma	NA	1.3††	0.21	0.16	NA	0.46††	NA	74	G1H1
	2 year	6 month			Right	5.7	≤0.1			Scrotal	No	Mature cystic teratoma	4.3††	0.63	3.7††	0.86	0.58††	0.54	1.06		
5¶	7 months§	1 year			Left	3.2	NA			Scrotal	No	Mature cystic teratoma	0.42	1.1††	0.38	0.34	0.34	0.25††	0.74	53	G1H1
6¶	7 months§	1 year			Left	NA	≤0.1			Scrotal	No	Mature cystic teratoma	0.33	0.95††	0.25	0.26	0.45	0.64††	1.42	50	G1H1

† Ratio to total volume of ipsilateral testis. ‡ Bilateral case. § Presented as testicular cyst and followed until solid mass became apparent. ¶ Dichorionic diamniotic twin. †† Affected side.

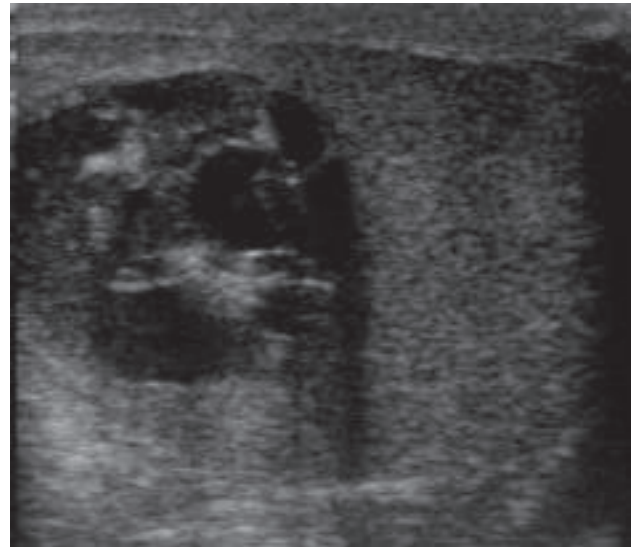


Fig. 1 Ultrasound image in Case 2, showing testicular lesion with heterogeneous echogenicity. In this case, the tumor was enucleated under cord clamping.

For 2 non-cystic lesions in older age, Cases 1 and 2 (Fig. 1), the spermatic cord was exposed via the transinguinal approach. The entire cord was clamped by a Fogerty vascular clamp, until the establishment of an intraoperative pathological diagnosis. In the other 5 tumors in 4 of the patients presenting a cystic lesion, the diagnosis of benign cystic teratoma was evident. Typically as in previous report,⁷ such tumor was initially presented as a cyst lesion, but tumorous component became evident when cyst shrunk away, in Cases 3, 5, and 6 (Fig. 2), within follow-up of 7–10 months. For such cystic tumors in infants, we essentially exposed the testis directly via the scrotal approach and enucleated the tumor without clamping the cord, with negligible bleeding because of the relatively limited vascularity at that age. In those cases, testis was approached transscrotally, except for the right side of Case 3, requiring ligation of the processus vaginalis for treating concomitant hydrocele.

The patients were followed up for 36–77 months (median 63.5). The residual testicular volume and absence of local recurrence were evaluated by ultrasound with a minimum 3-year follow-up. In all unilateral cases, there was over 60% of residual testicular volume in comparison to the contralateral side, irrespective of the initial tumor and cyst volume size or location. In Case 4, initial tumor and cyst volume size was 86% of affected testis volume, but remaining testicular volume was equivalent to contralateral side (Fig. 3). In 5 unilateral cases, we did not note radical change in testicular volume ratio to contralateral side during follow-up. The residual volume in 1 bilateral case, Case 3, was also within 2 standard deviation of age standard.⁸

Discussion

The novelty of this report was that residual testicular volume was precisely evaluated by ultrasound with a minimum 3-

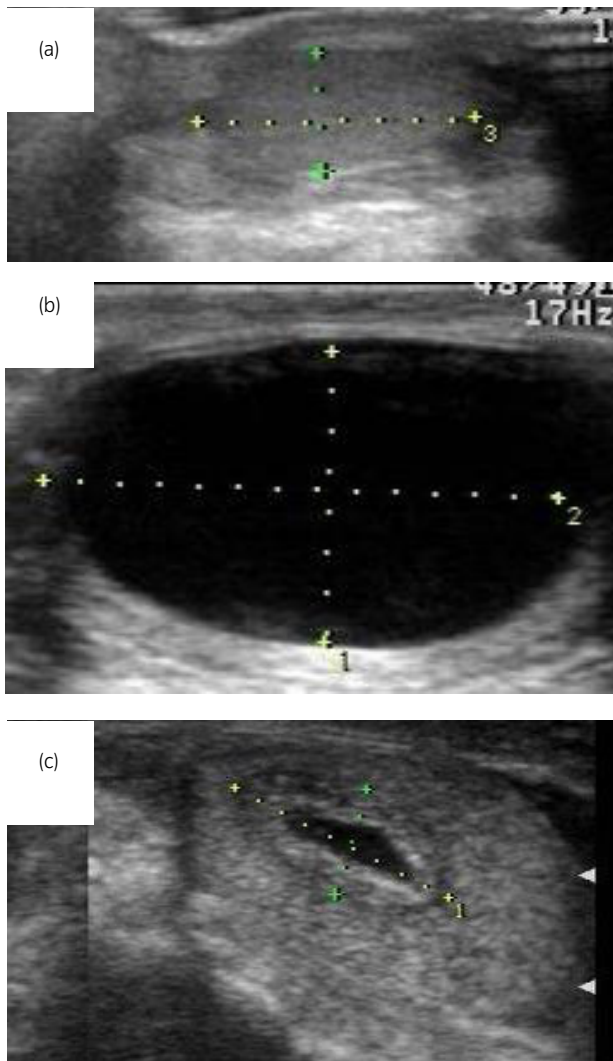


Fig. 2 Ultrasound image in Case 6, showing a natural history of pediatric cystic teratoma (size 15×6 mm). (a) At 5 months old, when the boy presented for hypospadias, no apparent lesion was noted. (b) At 7 months old, a cyst (size 19×11 mm) appeared in the center of the left testis displacing normal tissue. (c) At 17 months, a tumorous lesion (size 11×5 mm) appeared around the regressing cyst.

year follow-up, in a longer time frame than in previous literatures.^{9,10} In all unilateral cases, there was over 60% of residual testicular volume in comparison to the contralateral side, irrespective of the initial tumor volume including cyst or location. These results clearly indicated that the size of the cystic component could not be a reason for excluding tumor enucleation or justifying a total orchiectomy.¹⁰

It is well established transscrotal enucleation without vascular control is contraindicated for most testicular tumor, especially when malignancy is suspected. The 2 merits of cord clamping are the prevention of blood-borne metastasis and reduction in bleeding. We clamped the cord in 2 cases in older boys with noncystic lesion, in which malignancy was also suspected and vascularity was greater than that in infants. Though we enucleated them as prepubertal cases, we

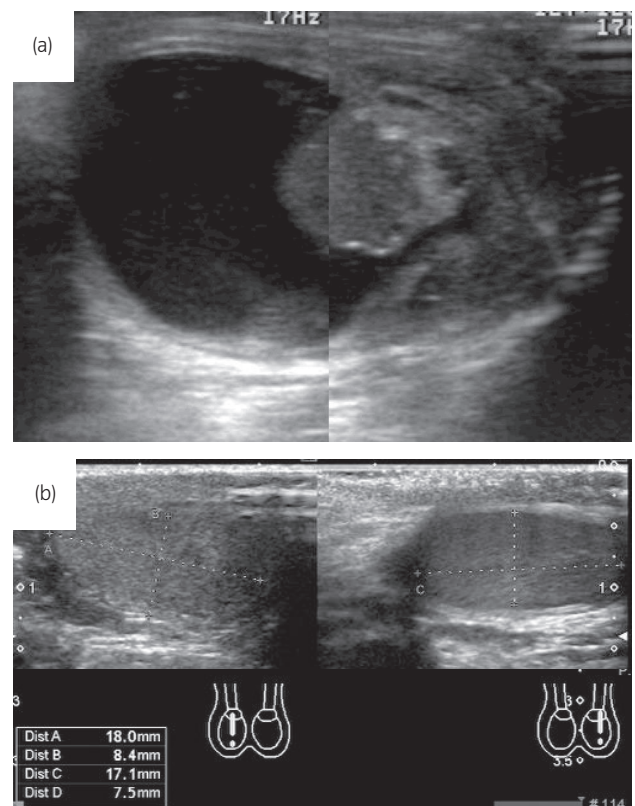


Fig. 3 Ultrasound image in Case 4. (a) Preoperative image showed tumor arising inside a cyst that displacing normal testicular tissue to periphery. (b) Postoperative follow-up image showed that affected right testis retained equivalent volume to contralateral testis.

would have chosen orchiectomy if they had been older. In such clinical decision, sonographic features are not helpful for excluding malignancy, except for cystic features, and clinical information such as age, solid tumor size, and AFP levels are more reliable.¹¹ Differentiating malignant tumor in children <1 year old is also problematic, because AFP level is elevated in normal children and one should be cautious in interpretation of the data. In clamped cases, ischemic time was about 60 min, but did not seem to have an effect on the residual testicular tissue volume on follow-up. It may not be surprising because testis can survive torsion ischemia up to 6 h.¹²

Nonetheless, when the preoperative diagnosis of cystic teratoma was established in infant cases with cystic lesion, there was little need to induce testicular ischemia in fine testicular vessels of this age group. If these cysts remained without sign of solid tumor, we would have continued following them up. In such cases, the transscrotal approach can achieve the surgical goal more easily and quickly.

The present series reporting 7 tumors is limited in size, but it is relatively large as a single surgeon series across several years and represents the outcome of a consistent surgical policy. Though not sufficient to lead to a definitive conclusion, such outcome confers insights not attainable from heterogeneous data sets deriving from multi-institutional studies³⁻⁵ or

from a single institute study from a longer time period, even though it would be possible to accumulate a larger number of cases.⁶ From the results of our cases, we may infer that clamped enucleation via the transinguinal approach may confer satisfactory preservation of the testicular tissue, but also unclamped enucleation via the transscrotal approach can also be a choice for preoperatively diagnosed benign cystic tumors in infants. Fertility data in these children are awaited for validating the impact of testis-sparing surgery in future.

Conclusions

Unclamped enucleation of testicular tumor via the transscrotal approach, which is contraindicated in most testicular tumors, can be a treatment choice exclusively for preoperatively diagnosed mature cystic teratoma in infants.

Conflict of interest

The authors declare no conflict of interest. This retrospective study was approved by the Institutional Review Board of Hyogo College of Medicine (Study number 3048). This retrospective study was disclosed in institutional website, and patients and their guardians had chance to opt out from the study. Because of retrospective nature of the study, this study was not registered.

Editorial Comment

Editorial comment to Preservation of testicular tissue after enucleation of pediatric mature teratoma: A case series of 7 testes in 6 children

Prepubertal testicular tumors (PTT) differ from those of postpubertal males in that benign lesions are more common. Testis-sparing surgery should be considered if preoperative evaluation, including alpha-fetoprotein (AFP) levels and ultrasonographic findings, suggests benign PTT with salvageable normal testicular parenchyma.¹


There have been only a few reports about the ultrasonographic evaluation of testicular volume after testis-sparing surgery.^{2,3} Kanematsu *et al.* evaluated the affected testicular volume by ultrasonography with a minimum 3-year follow-up and showed a residual testicular volume of >60% of the contralateral testicular volume.⁴ The authors should be congratulated on their work with a longer follow-up than that in the previous studies.

In addition, the authors concluded that transscrotal enucleation of the tumor without clamping the spermatic cord can be preferable for preoperatively diagnosed benign testicular

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tumors in infants.⁴ As I stated above, preoperative evaluation of AFP levels and ultrasonographic findings is particularly important. However, AFP levels in infants must be interpreted with caution because of its physiologically persistent elevation in children younger than 1 year of age. When we evaluate AFP levels in infants, the age-specific normal range reported by Tsuchida *et al.* may be helpful.⁵

Eiji Hisamatsu M.D. 

Department of Urology, Aichi Children's Health and Medical Center, Obu, Aichi, Japan
fpsfd412@yahoo.co.jp

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Conflict of interest

The author declares no conflict of interest.

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