Case Report

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

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Abbreviations & Acronyms
βhCG = beta human chorionic gonadotropin
AFP = alpha fetoprotein

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How to cite this article:
Kanematsu A, Yamamoto S. Preservation of testicular tissue after enucleation of pediatric mature teratoma: A case series of 7 testes in 6 children. IJU Case Rep. 2021; https://doi.org/10.1002/iju5.12328.

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Received 6 January 2021; accepted 15 May 2021.

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
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Introduction
Testis-sparing surgery has recently become the preferred treatment choice for pediatric benign tumors of the testis. 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. 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.
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

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**Discussion**

The novelty of this report was that residual testicular volume was precisely evaluated by ultrasound with a minimum 3-
year follow-up, in a longer time frame than in previous literatures.\textsuperscript{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.\textsuperscript{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 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.\textsuperscript{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.\textsuperscript{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 studies\textsuperscript{3–5} or
from a single institute study from a longer time period, even though it would be possible to accumulate a larger number of cases. 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.

References

1. Rushton HG, Belman AB, Sesterhenn I, Patterson K, Mostofi FK. Testicular sparing surgery for prepubertal teratoma of the testis: a clinical and pathological study. J. Urol. 1990; 144: 726–30.
2. Walsh C, Rushton HG. Diagnosis and management of teratomas and epidermoid cysts. Urol. Clin. North Am. 2000; 27: 509–18.
3. Valla JS. Valla JS for the Group D’Etude en Urologie Pediatrique. Testis-sparing surgery for benign testicular tumors in children. J. Urol. 2001; 165: 2280–3.
4. Ross JH, Rybicki L, Kay R. Clinical behavior and a contemporary management algorithm for prepubertal testis tumors: a summary of the Prepubertal Testis Tumor Registry. J. Urol. 2002; 168: 1675–8; discussion 8–9.
5. Gobel U, Haas R, Calaminus G et al. Testicular germ cell tumors in boys <10 years: results of the protocol MAHO 98 in respect to surgery and watch & wait strategy. Klin. Padiatr. 2013; 225: 296–302.
6. Hisamatsu E, Takagi S, Nakagawa Y et al. Prepubertal testicular tumors: a 20-year experience with 40 cases. Int. J. Urol. 2010; 17: 956–9.
7. Moriya K, Yamamoto S, Nakamura M et al. Spontaneous shrinkage of testicular teratoma in a prepubertal child. Urology 2017; 103: e13–e14.
8. Shukla AR, Woodard C, Carr MC et al. Experience with testis sparing surgery for testicular teratoma. J. Urol. 2004; 171: 161–3.
9. Patel AS, Coley BD, Jayanthi VR. Ultrasonography underestimates the volume of normal parenchyma in benign testicular masses. J. Urol. 2007; 178: 1730–2.
10. Chang MY, Shin JJ, Kim HG, Kim MJ, Lee MJ. Prepubertal testicular teratomas and epidermoid cysts: comparison of clinical and sonographic features. J. Ultrasound Med. 2015; 34: 1745–51.
11. Mellick LB, Sinex JE, Gibson RW, Mears K. A systematic review of testicle survival time after a torsion event. Pediatr. Emerg. Care 2019; 35: 821–5.