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# Bilateral Testicular Teratoma in Infancy: Report of a Rare Case Treated by Testis-Sparing Surgery



**Abstract**

Testicular tumors are rarely seen in childhood, with germ-cell tumors as the most common type. Teratoma is second only to yolk sac tumor in frequency. Bilateral testicular teratoma is extremely rare. Orchiectomy has been the standard treatment for most tumors of the testes in the past. However, in children, testis-sparing surgery has become the treatment of choice for benign lesions including teratoma.

A 10-month-old infant with synchronous bilateral testicular teratomas, one of which was disclosed by ultrasonography, is presented here. While the larger teratoma required unilateral orchiectomy, the smaller tumor in the contralateral testicle was enucleated by testis-sparing surgery. The follow-up at three years was uneventful with normal development of the testis and the boy.

Testicular teratoma may reside in both testicles without clinical symptoms. Ultrasound of both testes provides coherent preoperative diagnosis, allowing the surgeon to consider testicular-sparing procedures, thus preventing anorchia in these rare occurrences.

**Key words**
Testicular tumor · teratoma · testicular-sparing surgery · infancy

**Résumé**

Les tumeurs testiculaires, principalement de type germinal, sont exceptionnelles chez l'enfant. Le tératome est second en fréquence après le carcinome embryonnaire infantile. Les tératomes testiculaires bilatéraux sont extrêmement rares. Si pendant longtemps le traitement standard de la plupart des tumeurs testiculaires était l'orchidectomie, le traitement de choix chez les enfants atteints d'une tumeur testiculaire bénigne, dont le tératome, est une chirurgie conservatrice épargnant le testicule.

Un enfant de dix mois présentait deux tératomes testiculaires synchrones. Une orchidectomie était nécessaire pour exciser la première tumeur en raison de sa taille, mais une chirurgie conservatrice par énucléation était possible pour la deuxième, une découverte échographique au niveau du testicule controlatéral. A trois ans de suivi, l'enfant est en bonne santé avec un développement normal du testicule épargné.

Des tératomes testiculaires peuvent se développer au niveau des deux testicules sans manifestations cliniques. Grâce à l'ultrasonographie des deux testicules, un diagnostic préopératoire cohérent permet d'envisager une excision sans perte de testicule et d'éviter ainsi l'anorchidie dans les cas exceptionnels de tératomes bilatéraux.

**Mots-clés**
Tumeur testiculaire · tératome · chirurgie conservatrice du testicule · enfant



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**Resumen**

Los tumores testiculares son rara vez vistos en niños siendo los más comunes los de células germinales. De ellos, el teratoma es el más frecuente después del tumor del seno endodérmico. El teratoma testicular bilateral es extremadamente raro. La orquiectomía ha sido el tratamiento más común aplicado a los tumores testiculares en el pasado pero recientemente se ha desarrollado la posibilidad de cirugía conservadora del testículo como tratamiento de elección en las lesiones benignas incluyendo el teratoma.

Presentamos el caso de un niño de 10 meses con teratoma testicular bilateral sincrónico diagnosticado por ultrasonografía. Se practicó orquiectomía en el más grande pero pudo enuclearse el del testículo contralateral respetando parénquima testicular. En el seguimiento a tres años no hubo incidentes y se constató un desarrollo normal del testículo y del niño.

El teratoma testicular puede asentar en ambos testículos sin síntomas clínicos. La ultrasonografía de ambas gónadas nos permiten un diagnóstico preoperatorio coherente y la planificación de operaciones conservadoras del testículo para evitar la anorquia en estos raros casos.

**Palabras clave**

Tumor testicular · teratoma · cirugía conservadora · niños

**Zusammenfassung**

Hodentumore im Kindesalter sind sehr selten, wobei Keimzelltumore vor Dottersacktumoren und Teratomen am seltensten sind. Beidseitige Hodenteratome sind besonders ungewöhnlich. In der Vergangenheit galt die Orchidektomie als Methode der Wahl in der Behandlung von Hodentumoren. In jüngster Zeit wurden jedoch bei gutartigen Hodentumoren einschließlich Hodenteratomen gewebesparende Resektionsverfahren vermehrt angewandt.

Ein 10 Monate alter Knabe mit beidseitigem Hodenteratom wird vorgestellt. Bei dem größeren Teratom musste eine Orchidektomie durchgeführt werden, der kleinere kontralaterale Tumor konnte enukleiert werden. Drei Jahre nach dem Eingriff entwickelten sich die Hoden normal.

**Schlussfolgerung:** Hodenteratome können sich in beiden Hoden klinisch symptomlos entwickeln. Die Ultraschalldiagnostik erlaubt nicht nur die präoperative Diagnose, sondern ermöglicht auch Überlegungen hinsichtlich eines keimdrüsenerhaltenden Verfahren. Der vorliegende Fall ist publikationswürdig aufgrund seiner Seltenheit und der einseitigen partiellen Hodenresektion.

**Schlüsselwörter**

Hodenteratom beidseitig · hodengewebssparende Operationstechnik





## Introduction

Testicular tumors are rarely seen in childhood, accounting for 1 – 2% of all pediatric solid tumors (2). Germ-cell tumors are the most common type, comprising 60 – 75% of all prepubertal testicular tumors, although the percentage is lower compared to adult series (2). Testis is an uncommon site for teratomas, nevertheless testicular teratoma is second only to yolk-sac tumor within the germ-cell tumors (1,3). Bilateral testicular involvement by malignant germ-cell tumors is an uncommon entity, reported mostly in adult series. However, coexistence of teratomas in both testicles is extremely rare, with less than five reported cases to date (6,8,11). We report here an infant with synchronous bilateral testicular teratomas, successfully treated by unilateral orchiectomy, and contralateral tumor excision with testis-sparing surgery.

## Case Report

A 10-month-old boy was admitted with left testicular mass. The history revealed that he had been followed up with the diagnosis of hydrocele since he was 4 months old. In her first pregnancy three years previously, the mother reported a hydatiform mole, managed by methothrexate administration postoperatively. Her recent pregnancy was uneventful. The grandfather had undergone unilateral orchiectomy for an unknown mass at the age of nineteen.

On physical examination, a firm, mobile, painless left testicular mass, 2 cm in diameter, was prominent. The right testis was apparently normal. Routine biochemistry tests were normal except for a slight increase in serum alpha-fetoprotein (AFP) level (37.6 ng/ml, normal < 15 ng/ml). Both gray-scale and color Doppler ultrasonography revealed a sizable mass including cystic, solid, and calcified areas in the left testis, with a diameter of 19 × 13 mm (Fig. **1**). Unexpectedly, a second mass in the opposite testis was located caudally and anterolaterally which was smaller (6 × 3 mm) (Fig. **2**). The patient underwent surgical exploration through bilateral inguinal incisions. Both spermatic cords were occluded with non-crushing vessel loops before mobilization of the testes. Radical orchiectomy was performed on the left side which had inadequate normal tissue left. On the right, enucleation of the intratesticular mass was performed with preservation of surrounding normal testicular tissue. Histopathologically, both specimens were mature cystic teratomas. AFP level normalized postoperatively (8.2 ng/ml at 3 months postoperatively). There was no evidence of recurrent disease at three years follow-up, with normal echo pattern in the saved right testicle. Physical development was normal.

## Discussion

The clinical behavior of teratomas in children differs from that of adults. They have a benign clinical course in the prepubertal age, while metastases are frequently seen in adults. Only 2% of all testicular tumors appear in children, and about half are yolk sac tumors (2). Because of this and other data extrapolated from adults, the majority of these tumors have been managed with radical orchiectomy in the past. However, the development of the ultrasound technology made it much easier to demonstrate the cystic nature of teratomas, suggesting a preoperative diagnosis. Teratomas appear more as complex hypoechoic areas sur-

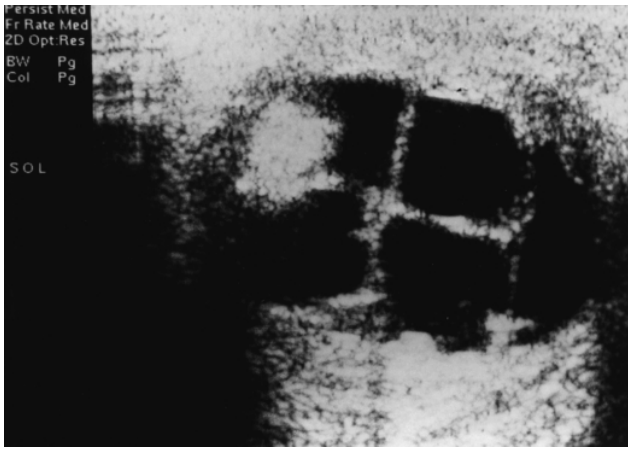


Fig. **1** Ultrasonography of the left testis shows a huge mass including cystic, solid, and calcified areas.

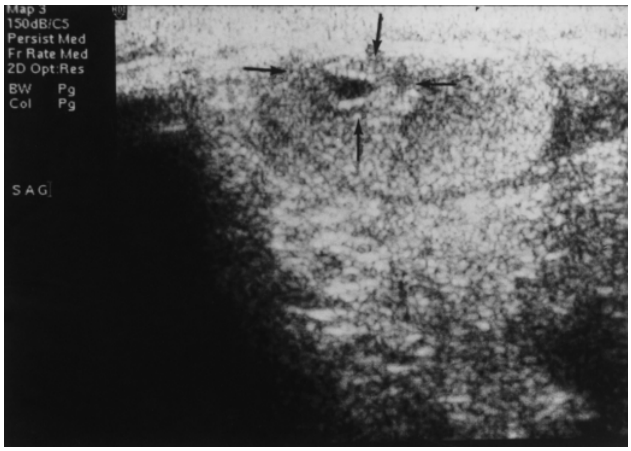


Fig. **2** Ultrasonography of the right testis demonstrates an intratesticular mass (arrows indicate the mass).

rounded by highly echogenic signals (4). Color Doppler ultrasound has been reported to be more effective than gray-scale ultrasound in detecting intratesticular neoplasms in the pediatric population (5).

If the preoperative evaluation suggests a benign intratesticular lesion, then a testis-sparing procedure can be considered (7,9). The management becomes more critical when both testes are involved as in our case. There have been no recurrences reported to date after testis-sparing surgery for benign lesions (7,9). Histologic diagnosis of teratoma is straightforward due to the characteristic features of the tumor. Rushton et al's review of 22 cases of prepubertal teratoma did not reveal any evidence of multifocal disease or carcinoma *in situ* of the adjacent testis (9).

Bilateral testicular teratoma, although very rarely reported in the literature, represents a critical situation where bilateral orchiectomy inevitably results in sterility, and deficiency of gonadal hormones (6). According to Brosman et al, there is often a delay of 7 months from first recognition of the scrotal swelling by the parents and initiation of treatment (1). This was typically observed in our case as well. The association of hydrocele with the larger tumor, which is a frequent feature of testicular tumors in children, might have led to a delay in presentation. The early diagnosis of contralateral intratesticular tumor by ultrasound prevented delay, and saved an extrasurgical procedure which would have been inevitable. Familial occurrence of testicular teratoma is rarely reported (10). In the presented case, the unknown testicular mass of the grandfather resected in his youth may suggest a familial occurrence.

Testicular teratoma may reside in both testicles without clinical symptoms. Ultrasound of both testes provides an important preoperative diagnosis, allowing the surgeon to consider testis-sparing procedures, thus preventing anorchia in these rare occurrences.

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