CASE REPORT

Paediatric benign cystic monodermal testicular teratoma

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SUMMARY

We present a case of a painless progressively increasing mass in the left upper testis in a 5-year-old boy, which was detected by his father performing testicular examination after observing asymmetry of the scrotum. Clinical examination, tumour markers, ultrasound scan including colour Doppler studies, and finally inguinal exploration and histopathology were helpful in establishing the diagnosis of benign cystic monodermal testicular teratoma. Testicular teratoma should be considered in the differential diagnosis of any child presenting with non-traumatic painless progressive scrotal swelling. Inguinal radical orchiectomy is the traditional gold standard treatment for benign testicular teratoma in children. Testis-sparing surgery should be reserved for smaller testicular tumours known to be benign.

BACKGROUND

Testicular tumours in children are rare, constituting about 1% of all paediatric solid tumours, and only 3% of all testicular tumours that arise in children.¹ Benign cystic testicular teratomas in prepubertal boys have not been reported to metastasise, whereas testicular teratomas in adults are associated with clinical metastasis in 60% cases.² We wish to report a case of an otherwise asymptomatic relatively small painless swelling to highlight the importance of testicular tumours at a young age as an important differential in scrotal swellings in children and awareness for early detection.

CASE PRESENTATION

A 5-year-old boy presented with a painless swelling of the left scrotum of 3 weeks duration. Examination revealed a firm, non-tender, non-transilluminable mass of about 1 cm diameter in the left testis. The right testis was normal.

INVESTIGATIONS

Serum α -fetoprotein (AFP) and β -human chorionic gonadotropin (β -HCG) levels were normal. Scrotal ultrasound scan showed a multiseptate heterogeneous mass compressing the remaining left testis; the right testis was normal (figure 1A). Colour Doppler ultrasound scan revealed normal vascularity around the swelling (figure 1B). On the longitudinal views of left testicular scan, a well-defined mass was seen occupying the upper pole of the left testis (figure 1C). A transverse scan confirmed a well-defined lesion with bright echoes suggestive of calcification (figure 1D). Abdominal ultrasound scan was normal.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included benign and possibly malignant testicular and paratesticular tumours.

TREATMENT

Exploration through a left inguinal incision revealed a firm mass, hard in places, within the left testis. A retrograde left orchiectomy was performed uneventfully. Histopathological examination revealed complete excision of a testicular epidermoid cyst (a benign cystic monodermal teratoma) occupying much of the testis, with calcification. Photomicrographs showed a well-circumscribed cyst with clear demarcation at the junction, compression of seminiferous tubules, and the cyst was lined by mature keratinising squamous epithelium. It had a wall of fibrous connective tissue, and within the cyst there were layers of compacted keratin which can sometimes give the cyst an 'onion ring' or 'target' appearance on scan (figure 2).

OUTCOME AND FOLLOW-UP

His postoperative recovery was uneventful and he was discharged home on the same day. He is asymptomatic, well, and there is no local recurrence at 3 months follow-up. The option of insertion of a cosmetic testicular prosthesis any time after adolescence has been discussed with the family.

DISCUSSION

Testicular tumours are much more common in adults than in children, the usual age at presentation being between 15 and 35 years. Paediatric testicular tumours are rare comprising only 1–2% of all paediatric solid tumours, and have an annual incidence of 1:100 000 for boys less than 15 years of age with a bimodal distribution of germ cell tumours with peaks below 2 years of age and in adolescence.

They are more common in Caucasians. The rate of testicular tumours being benign is higher in children (ranging from 38% to 74%) than in adults.³ In both adults and children even malignant testicular tumours are usually curable provided they are recognised early.

Testicular tumours in children can be benign or malignant, and they may arise from the non-germ cell derivatives to produce stromal tumours such as Leydig, Sertoli and gonadoblastoma tumours or from primordial totipotent germ cells which may develop into seminoma or embryonal carcinoma. Seminoma is primitive and unable to further

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Figure 1 (A) Transverse ultrasound scans of both testes together. (B) Colour Doppler study showing vascular nature of the left testicular lesion. (C and D) Longitudinal and transverse scan of left testis showing calcified polymorphic lesion.

differentiate, but embryonal carcinoma may differentiate into embryonic structures in the form of an epidermoid or dermoid cyst, a mature or an immature teratoma, or extraembryonic lesions leading to yolk sac tumour or a choriocarcinoma.

A painless progressive asymmetric lump in the testis is the commonest symptom of a testicular tumour. They may also

present with a feeling of 'heaviness' in the scrotum, a dull pain or discomfort in the testicle, groin, scrotum or abdomen.

Differential diagnosis includes epididymitis, torsion of the testis or of a testicular appendage and inguino-scrotal hernia. Approximately 10% of testicular tumours may have a secondary hydrocoele at presentation, so an underlying testicular tumour

Figure 2 Photomicrographs showing clear demarcation between cyst and the compressed testicular tissue and layers of compacted keratin within the cyst but no adnexa.

must always be excluded in any boy presenting with a hydrocoele. Occasionally a tumour in a testicle may lead to torsion of the testis, especially if it is undescended.

Elevated blood levels of tumour markers AFP, and less commonly β -HCG, increases clinical suspicion of the presence of specific testicular tumours and may help detect response to treatment and later recurrence. AFP is produced by the fetal yolk sac, liver and gastrointestinal tract and has a half-life of 5 days; its level is high in early infancy but falls to normal adult levels of <10 mg/mL by around 7–10 months of age. β -HCG is produced by embryonal carcinoma and mixed teratoma and may rarely be elevated in preadolescent tumours⁴; its half-life is 24 h.

Testicular self-examination is routinely recommended in adults who have previously suffered testicular maldescent and in cases of disordered sexual differentiation (DSD), but our case was brought to light by testicular self-examination by the father following observation of asymmetry of the scrotum. Given that the swelling was still not particularly obvious visually, our team appreciated his contribution towards his son's early detection.

Clinical examination with ultrasound scan including colour Doppler studies is good at diagnosing the testicular tumour but does not help in differentiating benign from malignant lesions. Anechoic cystic lesions suggest benign simple cyst, cystic dysplasia, teratoma or a benign granulosa cell tumour.⁵

Inguinal exploration and retrograde radical orchiectomy is the gold standard diagnostic and therapeutic procedure for testicular tumours in childhood. According to recent reports, testis-preserving surgery can be an option in the treatment of selective benign testicular tumours,⁶ but this is only appropriate when the mass can be easily separated from a worthwhile remnant of normal testis and there is already certainty that the pathology is benign. Intraoperative frozen section has its own well-documented pitfalls.

Once a final diagnosis has been made, the need for further investigation in the form of CT scans or MRI of abdomen and chest and other forms of treatments are decided on the merits

of each individual case.⁵ In conclusion, although testicular tumours are rare in children, a high index of suspicion may allow early detection and appropriate treatment.

Learning points

- ▶ Testicular teratoma should be considered in the differential diagnosis of any child presenting with non-traumatic painless progressive scrotal swelling.
- ▶ Inguinal radical orchiectomy is the traditional gold standard treatment for benign testicular teratomas in children.
- ▶ Testis-sparing surgery should be reserved for smaller testicular tumours known to be benign.

Contributors All the authors have made substantial contributions to the conception and design of this paper, search of literature, the acquisition, analysis and interpretation of the data, to drafting the article or revising it critically for important intellectual content, and to the final approval of the version to be published.

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