Republic); G Dahlquist (Sweden); A Green (Denmark); P Jarosz-Chobot (Poland); G Joner (Norway); D Michalkova (Slovakia); C Patterson (Northern Ireland); P Pozzilli (Italy, Lazio); AG Schranz (Malta); P Smail (Scotland); M Songini (Italy, Sardinia); B Urbonaite (Lithuania); E Wadsworth (British Paediatric Survey, UK).

Funding: The European diabetes (EURODIAB) research network is supported by the European Commission medical research programme (contracts BMH1-CT92-0043 and BMH4-CT96-0577).

Competing interests: None declared.

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*(Accepted 4 May 1999)*

# Testicular neoplasia in cryptorchid boys at primary surgery: case series

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*BMJ* 1999;319:888–9

Cryptorchidism is associated with testicular cancer; the lifetime risk of 2-3% is about four times higher than in the general population.[1 2] Some groups of cryptorchid patients may have an especially high risk of testicular cancer.[3] Testicular carcinoma in situ is a well described histological pattern that precedes germ cell tumours.[1 4] We investigated whether it is possible at primary surgery to identify cryptorchid boys who have testicular neoplasia and therefore are at high risk of testicular cancer.

## Method and results

We examined 1535 consecutive specimens of testicular tissue that were obtained from undescended testes at surgery for cryptorchidism in 1249 boys between 1971 and 1998. Previous reports have described 1026 of the biopsies in detail.[2 4] No patient had fallopian tubes or a uterus.

The table shows the total occurrence of testicular neoplasia at surgery for cryptorchidism. There was one case of invasive germ cell tumour, six cases of testicular carcinoma in situ, and one Sertoli cell tumour. Of the eight testes with neoplasia from seven patients, three neoplasms were diagnosed in intra-abdominal testes (cases 1-3), four occurred in three boys with abnormal external genitalia other than cryptorchidism (cases 4-6), and two were diagnosed in boys with known abnormal karyotype (cases 3 and 7).

All the case reports were reviewed. In the 97 boys (124 specimens) with intra-abdominal testes, three had known abnormal karyotype; 46,XY/47,XYY (case 3), 46,XYdel(11p), and 46,XY,13/20 unbalanced translocation; five had abnormal external genitalia, two with hypospadias, one with epispadias, and two with small penis and scrotum. Furthermore, 28 patients (38 specimens) had abnormal external genitalia but no intra-abdominal testes: 14 with hypospadias, two with epispadias, two with some ambiguity of the external genitalia (cases 4 and 5), two with hypoplastic scrotum, and eight with small penis and scrotum, of whom four had Kallmann's syndrome and one had testicular neoplasia (case 6). Moreover, 10 patients (14 specimens) had known abnormal karyotype: seven with 47,XXY;

Testicular neoplasia in eight undescended testes from seven cryptorchid boys, among 1249 patients who at median age 12.0 years (range 0.1-18.9 years) underwent surgery for cryptorchidism with examination of 1535 specimens of testicular tissue from undescended testes

| Case No | Age at surgery for cryptorchidism | Anatomical position of testes | Record of testicular neoplasia and finding in contralateral testis | Characteristics |
|---|---|---|---|---|
| 1 | 13.3 (right) | Intra-abdominal | Germ cell hypoplasia | Intra-abdominal testes, 46,XY; genitalia not abnormal |
| | 13.3 (left) | Intra-abdominal | Carcinoma in situ testis | |
| 2 | 5.4 (right) | Scrotum | Parents wanted no biopsy | Intra-abdominal testis, 46,XY; genitalia not abnormal |
| | 5.4 (left) | Intra-abdominal | Carcinoma in situ testis | |
| 3 | 7.1 (right) | Intra-abdominal | Large cell calcifying Sertoli cell tumor of testis | Intra-abdominal testes and abnormal karyotype; 46,XY/47,XYY; external genitalia not abnormal |
| | 7.1 (left) | Intra-abdominal | No germ cells pattern | |
| 4 | 10.2 (right) | External inguinal ring | Carcinoma in situ testis | Abnormal external genitalia; small penis, vaginal pouch, bifid scrotum, 46,XY[1] |
| | 10.6 (left) | External inguinal ring | Carcinoma in situ testis | |
| 5 | 10.9 (right) | External inguinal ring | Carcinoma in situ testis | Abnormal external genitalia; small penis and partially bifid hypoplastic scrotum[4] |
| | 10.8 (left) | External inguinal ring | Germ cell hypoplasia | |
| 6 | 18.6 (right) | Inguinal canal | Seminoma | Abnormal external genitalia; small penis and hypoplastic scrotum, 46,XY[4] |
| | 18.6 (left) | External inguinal ring | Germ cell hypoplasia | |
| 7 | 15.4 (right) | Inguinal canal | Carcinoma in situ testis | Abnormal karyotype; 45,X/46,XY[4] |
| | 15.4 (left) | Scrotum* | Germ cell hypoplasia | |

In total, the risk of testicular neoplasia at surgery for cryptorchidism in childhood was: 7/1249=0.56% (exact 95% confidence interval 0.28% to 1.15%; calculated by solving the exact binomial equations iteratively) per cryptorchid boy, and 8/1535=0.52% (0.27% to1.02%) per testicular specimen from an undescended testis.
*This biopsy from a scrotal testis was not included in the material of 1535 specimens of testicular tissue from undescended testes.

one with 45,X/46,XY (case 7); one with 46,XX; and one with 47,XYY.

At surgery for cryptorchidism the risk of testicular neoplasia was 7/135 (5.2%) in patients with intra-abdominal testis, abnormal external genitalia other than cryptorchidism, or diagnosed abnormal karyotype. In contrast, no case of testicular neoplasia occurred in 1114 patients without these characteristics. The figures are significantly different (Fischer's exact test, $P < 0.001$).

At surgery the risk of testicular neoplasia was 4/286 (1.4%) in the 286 patients operated on for bilateral cryptorchidism, which is not significantly different from 3/963 (0.3%) in the 963 patients with unilateral cryptorchidism ($P = 0.10$). However, intra-abdominal testis, abnormal external genitalia, or abnormal karyotype were reported in 42 (14.7%) patients operated on for bilateral cryptorchidism, compared with 93 (9.7%) patients with unilateral cryptorchidism ($P < 0.05$).

## Comment

All seven boys with testicular neoplasia at surgery for cryptorchidism had intra-abdominal testis, abnormal external genitalia, or known abnormal karyotype. No case of testicular neoplasia was found in patients without these characteristics. To our knowledge, this information is new, but it is not inconsistent with the literature.[1 4 5] At operation for cryptorchidism the surgeon may elect to request a biopsy of testes with a high risk of neoplasia. This bias may explain the conflicting interpretation of our previously published results[2] and the results published by Swerdlow et al.[3] In clinical practice, we recommend a testicular biopsy at surgery for cryptorchidism if the boy has intra-abdominal testis, abnormal external genitalia, or known abnormal karyotype. These abnormalities were more common in patients with bilateral than with unilateral cryptorchism. The suggested procedure may diagnose testicular neoplasia at surgery for cryptorchidism and some patients may be treated for testicular neoplasia before invasive cancer develops.

Contributors: JV verified the pathological data and contributed to interpretation and reporting. HM contributed to interpretation and reporting. JT verified the clinical data and contributed to study design, interpretation, and reporting. DC took the initiative in this study, verified the pathological and clinical data, and was responsible for interpreting the results and drafting the paper. All authors are guarantors.

Funding: No specific funding.

Competing interests: None declared.

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*(Accepted 12 May 1999)*

*A lesson learnt*

## An inspirational teacher

It was my first experience of terminal care—I was a trainee, very different then, the apprenticeship. Starting off with hand held, then the reins lengthening, and finally Jimmy said that he would cover from a weekend away fishing. We were not on call, but his patient with breast cancer was almost certainly going to die over the weekend, so would I "do the needful?"

I know the theory, but not a lot of the practice—visit the family a few times with Jimmy beforehand, try and adopt the same air of breezy confidence and support, give them my home telephone number, arrange to visit several times daily. I cannot remember her name, but I can see her lying in bed, licking dry lips, her husband attentive, giving sips of water off a spoon, the district nurses busy with skin care, adjusting sheepskins, rubbing heels, turning gently, but there was obviously pain and distress. I increase the diamorphine, she's more settled, but still breathing regularly. Will it happen very suddenly? I'd better not go out in case they telephone. It's a glorious June day and we live in a flat. I don't question that I'm doing this on my weekend off. I know if Jimmy was here that's what he would be doing and I'm flattered that he trusts me to do it.

I'm in and out like a yo-yo all the weekend—she's unconscious but not dead, her breathing noisy and laboured. Jimmy comes back on Sunday night and we decide to increase the diamorphine again. She dies a few hours later and that's my weekend.

What's the learning in this? It's more about training than palliative care. Dr Jimmy Ledingham died last year (obituary, 9 January 1999, p 129) while visiting an ex-patient. I've been thinking about him a lot and what he meant to me and his patients—how he related to the patient, to her husband, to the nurses, to me. He was so down to earth and cheerful but hugely supportive and so obviously capable, knowing what symptoms were most distressing, knowing when people needed to talk, trusting me to take over.

Coming back, still in his fishing gear, tweeds, woollen jumper—utterly dependable. I wanted to be the same, yet as time went on I wanted to be as good as a mum as I was as a doctor. It was difficult to be both, particularly the same kind of doctor as Jimmy.

But terminal care does not happen very often: the intensive part may be just a few days, and the rewards are huge—feeling you're doing your job well, cementing relationships with the family. It's what we would all want for our own families. It is so important to pass that on to those we teach. I'm trying to do that this week as my registrar and I share the care of our terminally ill patient.

Jimmy was an inspirational doctor and man, and I miss him terribly. His legacy to me, as a trainer myself now, is to remind me of the importance of teaching by example, which matters as much as, if not more than, anything that happens in a tutorial.

Alison Douglas, *general practitioner and trainer, Aberdeen*

We welcome articles up to 600 words on topics such as *A memorable patient, A paper that changed my practice, My most unfortunate mistake,* or any other piece conveying instruction, pathos, or humour. If possible the article should be supplied on a disk. Permission is needed from the patient or a relative if an identifiable patient is referred to. We also welcome contributions for "Endpieces," consisting of quotations of up to 80 words (but most are considerably shorter) from any source, ancient or modern, which have appealed to the reader.