Polyorchidism

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Summary: Polyorchidism is an unusual abnormality of the genital tract in which supernumerary testes are present, usually within the scrotum. We report a recently encountered case, discuss the aetiology of the condition and suggest a course of management when encountered at operation.

Introduction

Polyorchidism is an unusual abnormality of the genital tract in which supernumerary testes are present, usually within the scrotum. To date there have been only 40 cases reported in the English literature. In only eight such cases has histologically normal spermatogenesis in the accessory testis been described and in only one have both testicular masses been of equal size. We report a case where spermatogenesis in both testicular masses was normal and where both masses were of equal size, discuss the aetiology of the condition and suggest a course of management when encountered at operation.

Case report

A 17 year old male presented with a two-year history of a left-sided scrotal swelling. There were no other symptoms attributable to the genitourinary tract. Clinical examination of his scrotum revealed a normal right testis, epididymis and spermatic cord. The left side of the scrotum however, contained two distinct swellings; one was thought to be a hypoplastic testis and the other to be a cyst within the spermatic cord. Exploration of the left side of the scrotum was undertaken.

At operation two distinct testicular masses of equal size were found within the left side of the scrotum with a small associated hydrocoele. These masses were attached to a common epididymis, and the lower one was served by a single vas deferens (Figure 1). The upper of the two masses was excised and the lower biopsied.

Histological examination of both the excised upper testis and the biopsy of the lower testis revealed

normal testicular tubular architecture with normal maturation of germ cells and numerous spermatids. The post-operative course and subsequent follow-up of the patient was unremarkable.

Discussion

The first histologically documented case of polyorchidism was reported by Lane in 1895¹ but there are numerous unsubstantiated reports in history of men boasting up to six testes and associated superhuman fecundity. Forty histologically proven cases have since been reported in the literature, although only eight

Figure 1 Both testes are of equal size and share a common epididymis and vas deferens.

Figure 2 Classification of reported cases of polyorchidism (including current case) based on site of accessory testis.

have reported normal spermatogenesis in the accessory testis and only one exhibited duplicated testes of equal size.²

Two theories exist as to the cause of such testicular duplication. Longitudinal duplication of the genital ridge would result in the development of two separate testes. Only the lateral mass would have any connection with the developing mesonephric duct which ultimately forms the epididymis and vas deferens. In such circumstances it might be expected that the total volume of both testes would exceed the expected volume of one alone. Transverse division of the genital ridge during development would result in a number of possible anatomical anomalies depending on the level of the abnormal division. A common vas and epididymis shared by both testicular masses is the most frequent situation, but duplication of one or both of these structures in addition to duplication of the testis has been reported. Division through the mesonephric duct in addition to division through the genital ridge would result in only one of the testicular masses being connected to a vas deferens although both would have separate epididymii. The totally isolated testis usually lacks active spermatogenesis. In transverse divisions the total mass of both testes together is usually not greater than the equivalent of one normal sized testis.^{2,3}

Hancock and colleagues classified the reported cases to date in two ways.⁴ The site of the accessory testicular mass was used in the first classification to divide the cases into three groups; scrotal, inguinal and retroperitoneal (Figure 2). In the second classification the detailed anatomy of the testes, epididymii and vas was used to distinguish the groups (Figure 3). Of the 36 cases reviewed the abnormality occurred on the right side in 12 patients and on the left side in 24.

The clinical presentations of these patients varied. The commonest presentations were of two masses within the hemiscrotum (scrotal group), inguinal swelling (due to testicular mass or associated indirect inguinal hernia), undescended testis (inguinal and

Figure 3 Classification of reported cases of polyorchidism (including current case) based on anatomy of testes, epididymii and vas deferens.

retroperitoneal groups) and pain from torsion of the abnormal accessory testis. Some patients were found to have associated hydrocoele or varicoceles. There have been at least two reports of testicular tumours developing in polyorchid patients (both in the inguinal group) suggesting that the incidence of malignant change in these abnormal testes is higher than would be accounted for by cryptorchidism alone.^{5,6}

There has been some debate as to how best to manage these patients. It is generally accepted that cryptorchid testes should be brought down into the scrotum in early life to ensure as normal function as possible, both hormonally and spermatogeneically, to allow regular examination of the testis to detect malignant change and to prevent any psychological trauma that the absence of a testis might cause. On this basis it would seem reasonable to undertake orchidopexy in the cryptorchid cases of polyorchidism. There is virtually no evidence available to suggest that orchidopexy reduces the risk of malignant change in cryptorchid testes.⁷ The advantage of orchidopexy would seem to be that easy examination of the at-risk organ is facilitated. The age at which orchidopexy seems to have little effect on the subsequent risk of testicular cancer.⁸

The high incidence of torsion in the accessory testis would support early orchidopexy. The fact that only 1 out of 40 reported cases have demonstrated histological evidence of normal spermatogenesis, would perhaps lend support to the opinion that the abnormal tissue should be excised, particularly in view of the high theoretical risk of malignant change. Some

authors, however, suggest that all gonadal tissue should be left in the scrotum to maximise potential fertility.⁹ This seems illogical given that most of the accessory testes are histologically abnormal, and as such would not be expected to contribute to fertility.

We suggest that the least anatomically normal testis should be excised (namely that without attachment to

an epididymis and vas) and that the remaining testis should be submitted to biopsy and surgically fixed in the scrotum. The absence of spermatogenesis in, or an abnormal histological report on, the biopsied testis should indicate the need for increased vigilance in follow-up of patients at higher risk of developing testicular malignancy.

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