Case – Obstructive azoospermia secondary to bilateral hydroceles

Matthew Mancuso, Phil Vu Bach

Division of Urology, Department of Surgery, University of Alberta, Edmonton, AB, Canada

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INTRODUCTION

Clinical infertility is the inability of partners to conceive after 12 months of unprotected intercourse.[1] It is estimated that male factor is solely responsible for 20–30% of infertility, although male factor can be contributory in up to 50% of infertility cases.[2] A thorough history, physical exam, and investigations such as semen analysis are critical in the evaluation of male infertility.[3]

Azoospermia, or the complete absence of sperm in the ejaculate, occurs in approximately 1% of all men and is typically broken down into non-obstructive azoospermia (NOA) and obstructive azoospermia (OA).[4] NOA stems from pre-testicular or testicular failure, whereas OA results from post-testicular blockage of the male reproductive tract, with normal spermatogenesis maintained within the testis.[5] Testicular volumes and follicle-stimulating hormone (FSH) levels are typically used to help differentiate NOA from OA.[6]

Common etiologies for OA include vasectomy, scrotal surgery, congenital absence of the vasa deferens, and epididymitis.[7] Since spermatogenesis is maintained in OA, the management of OA consists of either reestablishing continuity of the male reproductive tract (e.g., vasovasostomy) or sperm retrieval techniques for artificial reproductive technologies (ART).[8] Numerous sperm retrieval techniques exist and can involve microsurgical, percutaneous, or open approaches to retrieve sperm from the testis or epididymis.[9]

Within the scrotum, the testes are surrounded by the tunica vaginalis. When fluid accumulates between the visceral and parietal layers of the tunica vaginalis, a hydrocele forms. Hydroceles are common findings and typically cause minimal clinical concern.[10]

The following case presents the first description in the literature of a patient presenting with OA secondary to very large bilateral hydroceles, with bilateral hydrocele repair performed to facilitate testicular sperm retrieval instead resulting in complete normalization of ejaculate semen parameters.

CASE REPORT

Presentation and workup

The patient is a 43-year-old man referred to urology after 12 months of secondary infertility and azoospermia. He and his partner (40-year-old G3P1L1SA2 female) had a seven-year-old child naturally conceived without difficulties and two naturally conceived miscarriages. He presented with an unremarkable medical history, with no contributory etiologies identifiable for his azoospermia presentation; no prior cryptorchidism, genital or pelvic surgery, urinary tract or sexually transmitted infections, smoking, medical or family history of significance.

Physical exam findings at consultation included palpable vasa deferens bilaterally and large 10 cm hydroceles bilaterally that prevented palpation of his testes. Investigations revealed a normal hormone profile and azoospermia on two separate semen analyses (Tables 1–3). Since testicular palpation was not possible, scrotal ultrasound was performed, which revealed normal bilateral testes and 9.5 cm right and 9.6 cm left hydroceles. The sonographer was unable to visualize the epididymides but confirmed a lack of varicoceles.

While the exact time of hydrocele onset after their naturally conceived child was unclear, a scrotal ultrasound two years prior to presentation demonstrated 6 cm bilateral hydroceles. Genetic investigations confirmed a normal karyotype with no Y-chromosome microdeletions. Given his prior proven fertility, normal volume testes, normal FSH levels congruent with OA, and with his partner's advanced age, surgical sperm retrieval for use with in vitro fertilization (IVF) was recommended. To facilitate surgical sperm retrieval for cryopreservation, bilateral hydrocelectomies were organized.

Table 1. Initial hormone panel at time of consultation

Investigation	Value	Normal
Testosterone, total am	12.9 nmol/L	10.3–29.5 nmol/L
Estradiol	48 pmol/L	<200 pmol/L
FSH	4.8 IU/L	<7 IU/L
LH	3.3 IU/L	<12 IU/L
Prolactin	7.7 ug/L	<21 ug/L

FSH: follicle-stimulating hormone; LH: luteinizing hormone.

Table 2. Initial semen analysis

Parameter	Value
Semen volume	3.0 mL
Sperm count	0
Semen pH	8.1

Table 3. Repeat semen analysis

Parameter	Value
Semen volume	2.4 mL
Sperm count	0
Semen pH	7.9

Operative details

An uncomplicated, standard bilateral hydrocelectomy was performed; briefly, a median raphe incision, blunt and electrocautery dissection, delivery and drainage of straw-colored hydroceles, excision of redundant hydrocele sacs, and Jaboulay oversewing were the techniques used bilaterally. The epididymis did not exhibit gross dilation nor any transition points in keeping with focal obstruction, which may suggest compression of the epididymal tubules from the elevated intrascrotal pressures. The supero-anterior portion of the right testis was positioned just below the surgical incision for ease of accessibility in a subsequent testicular sperm retrieval.

Postoperative details

The patient was seen four weeks postoperatively with no complications or concerns. Benign hydroceles sacs were reported on pathology. Semen analysis and hormone panel were then repeated at four months postoperatively, which showed full return of sperm to his ejaculate (Table 4). Given that his semen parameters were back to normal range sufficient for natural conception or any form of ART, plans for surgical sperm retrieval were abandoned. He and his partner decided against ART and opted to continue trying for natural conception.

Table 4. Post-hydrocelectomy semen analysis

Parameter	Value
Semen volume	3.5 mL
Sperm concentration	27 000 000/mL
Motility	44%
Morphology	3%
Semen pH	7.9

DISCUSSION

Within the existing literature, the causes for OA are fairly well-described. To our knowledge, this is the first case that describes bilateral hydroceles as a cause of OA. While iatrogenic injury during hydrocelectomy is a known potential cause of OA, this case demonstrates how the hydroceles themselves can affect sperm transport and how repair of large hydroceles may have a therapeutic benefit in the management of OA when no other etiologies are identified.

Consideration was given to have his hydrocelectomies and surgical sperm retrieval done simultaneously, but logistical issues at our institution prevented concurrent procedures. Fortunately, the return of sperm to his ejaculate post-hydrocelectomies meant that no additional surgery was needed. Therefore, we stress the importance of repeating the semen analysis after operative intervention of the reproductive tract prior to further intervention.

The smooth muscle layers of the vasa consist of a middle circumferential layer surrounded by two longitudinal layers that function together to coordinate the transport of sperm during ejaculation.[11,12] While human data is lacking, the robustness of the vas deferens in animal models in response to various degrees of iatrogenic surgical injury has been studied. When subjected to 30 seconds of digital compression, mild to moderate inflammation with occasional focal narrowing of the rat vasal lumen was noted on interval histologic examination of the vas six weeks later; however, patency was maintained in all cohort rats.[13]

Guinea pig studies have described intraluminal hydrostatic pressures in the seminiferous tubule to range between 2–4 cmH_2O and in the epididymis ranging from 2–28 cmH_2O.[14] Intrascrotal pressures up

to 21 cmH_2O from hydroceles in humans have been reported compared to contralateral (control) measurements of 0 cmH_2O.[15] The extrinsic mechanical force exerted by a hydrocele may therefore affect the vas deferens, epididymis, and/or seminiferous tubules.

Despite this significant increase in extrinsic intrascrotal pressure exerted by a hydrocele, which may overcome the resting intraluminal hydrostatic pressures of the sperm outflow tract, it is widely assumed that hydroceles independently are not risk factors for OA. Histologic studies have previously confirmed preservation of spermatogenesis in men with large hydroceles.[14]

In our case, we suspect that large, bilateral hydroceles may have generated sufficient pressures on the sperm outflow tract, ultimately causing OA. Upon operative repair, this obstructive force was sufficiently relieved to allow sperm transport through the seminiferous tubules, epididymides, and vasa deferens back into the ejaculate. Further investigations on the capacity of the seminiferous tubules, epididymis, and vas deferens to withstand extrinsic compression forces, as well as the ability of large hydroceles to generate intrascrotal pressure, would be helpful in understanding quantitatively how hydroceles can have a negative impact on male fertility.

CONCLUSIONS

This is the first reported case of a man with secondary infertility and OA due to large bilateral hydroceles. While iatrogenic injury during scrotal surgery is a known risk factor for OA, this case shows how bilateral hydrocelectomies can be therapeutic in relieving OA.

COMPETING INTERESTS: The authors do not report any competing personal or financial interests related to this work.

This paper has been peer reviewed.

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CORRESPONDENCE: Dr. Matthew Mancuso, Division of Urology, Department of Surgery, University of Alberta, Edmonton, AB, Canada; mmancuso@ualberta.ca