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Infection—Stones

EPIDIDYMITIS IN INFANTS AND BOYS: UNDERLYING UROGENITAL ANOMALIES AND EFFICACY OF IMAGING MODALITIES

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ABSTRACT

Of 47 children diagnosed as having epididymitis between 1975 and 1985, 8 of 17 prepubertal patients (47 per cent, or 17 per cent of the total group), including 3 of 4 infants (75 per cent), had an underlying urogenital anomaly. Thus, patients with epididymitis secondary to an underlying anomaly present at an earlier age. The common finding in patients with underlying anomalies was a pathological connection between the urinary tract and the genital duct system or bowel.

Coliform urinary infections predominated in patients with underlying anomalies (4 of 6). Although a positive urine culture suggested an underlying anomaly (positive predictive value 0.60), a negative culture virtually ruled out an anomaly (negative predictive value 1.00).

Of the 8 patients with underlying anomalies 6 underwent voiding cystourethrography and 6 underwent excretory urography, and the diagnosis was established in 6 (100 per cent) and 4 (67 per cent), respectively. Thus, voiding cystourethrography has the greatest diagnostic yield. Any prepubertal patient with epididymitis merits a complete urological evaluation, including a urine culture, voiding cystourethrography and excretory urography to rule out an underlying urogenital anomaly, which often is amenable to an operation. Surgery often involves severance of the pathological urogenital or urinary-fecal connection and it was successful in 7 of our 8 patients. (*J. Urol., part 2, 138: 1100–1103, 1987*)

Epididymitis is a rare but important diagnostic entity in the infant or child who presents with acute scrotal inflammation. Clinically, epididymitis is difficult to differentiate from other noninflammatory scrotal processes, including torsion of the spermatic cord, appendix testes or appendix epididymis, hernia, hydrocele or tumor. Scrotal exploration often is performed and the diagnosis of epididymitis is made by the process of exclusion of other entities.

We reviewed a 10-year experience with epididymitis at a busy pediatric center with particular focus on the types of underlying urogenital anomalies that may predispose and contribute to inflammation of the epididymis in childhood. The efficacy of diagnostic imaging modalities used to evaluate these cases is addressed and the use of clinical or laboratory factors to predict that population with underlying urogenital anomalies is determined.

MATERIALS AND METHODS

The records of all patients who presented to our hospital with the diagnosis of epididymitis between 1975 and 1985 were reviewed. All available inpatient and outpatient data were studied, as well as all available radiographs. Factors that were reviewed included age, clinical presentation, operative findings if performed, radiographic evaluation, laboratory tests (white blood count, urinalysis and urine culture) treatment and followup.

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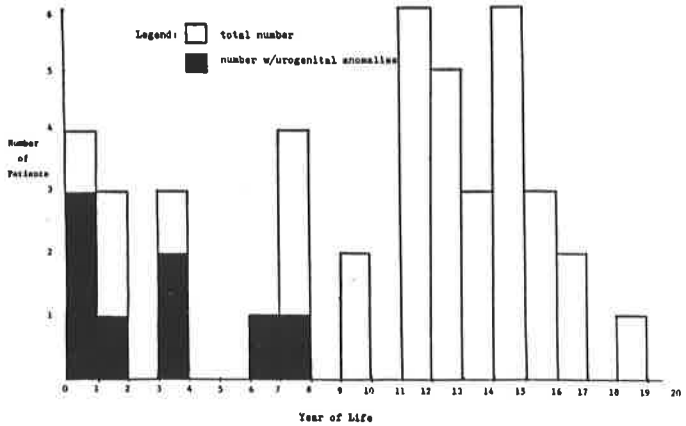
RESULTS

Between 1975 and 1985, 47 patients were diagnosed with acute epididymitis (4.7 patients per year). Patient age at presentation ranged from birth to 19 years (mean age 12 years). Of the patients 40 per cent (17 of 43) were prepubertal (4 presented during first year of life) and 60 per cent (26 of 43) were postpubertal, with puberty defined arbitrarily as a patient age of 11 years. In 4 patients the age was unavailable. Most of the patients presented with scrotal erythema, swelling and pain. Epididymitis was diagnosed at scrotal exploration in 13 of the 47 patients (28 per cent).



Age distribution versus distribution of underlying urogenital anomalies.

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DISCUSSION

Dr. E. Reda. I compliment the authors for pointing out the various situations in which bladder mucosa can be used as either a patch graft or a tubularized graft. It is interesting that this type of graft was advocated in 1947 and 40 years later we see a resurgence of enthusiasm for its use. One of the most important messages in these excellent series of patients and perhaps what many of us now take for granted is that the better results represent not only the type of graft itself but a greater awareness of the importance of fine technique, improved small suture material, optical magnification, appropriate antibiotics, nonirritating and malleable polyethylene stents, and carefully constructed compression dressings.

Koyle and Ehrlich should be congratulated on their outstanding results, particularly in 7 hypospadiac cripples who had experienced previously multiple failed operations. Most of us began to use bladder mucosa when we thought we had little to lose in attempting a new technique. However, these authors also report on 3 patients in whom they used bladder mucosal grafts as a primary procedure in preference to other sources of extragenital skin. Clearly, the trend presented by these presentations points toward the use of bladder mucosa rather than extragenital skin because of its more favorable characteristics. I am impressed by the lack of meatal problems, since this has been a recurrent concern in the series of Hendren in 1986 as well as my own experience during the last 2 years.

It should be noted that Ransley and associates encountered excess protruding bladder mucosa at the meatus in 50 per cent of the cases associated with total urethral replacement, which is in accord with our series and that of Hendren. Mr. Ransley has attempted to use silver nitrate but as I understand it, he is less than pleased with the eventual outcome and he believes that most if not all will require excision. Although excision has proved to be a simple solution to this difficulty, since it is performed on an ambulatory basis, it represents a nuisance in that the extruded mucosa bleeds easily, tends to stick together, produces splaying of the stream and is unsightly cosmetically. In the series of Koyle and Ehrlich I would have thought that there would be a higher incidence of this problem, since it was stated that an extra 10 per cent of graft length is harvested regularly to allow for subsequent retraction.

The complication that neither paper addresses, which we have encountered in 3 of our 35 cases, is ballooning of the urethra caused by overestimation of graft width. We have the distinct impression that free bladder mucosa grafts contract to a much lesser extent than either penile or extragenital skin. We are well aware of the adage that states that it is better to make a graft wider than too narrow. Someone always says that slow flow is better than no flow at all but it is even better to make the graft just right. Previously, we overcompensated by 40 per cent but now we are doing what Koyle and Ehrlich suggest. Ransley and associates address yet another important application for free bladder mucosa other than that of the tubularized graft. They used bladder mucosa as a patch graft to advantage in cases of urethral strictures.

Dr. Frank Motrell. Mr. Ransley, is it possible to use a pedicle flap from that small distal margin rather than another free graft and perhaps improve your results? I know it cannot be done in some complicated repeat cases but it might be an alternative when possible.

Dr. Thomas H. Bartholomew. Practice does not make perfect with hypospadias repairs. How tightly should one stretch the bladder mucosa before measurements are made?

Dr. Emilio M. Quesada. We do not have much experience in the use of bladder mucosa but we have performed total replacement in 8 cases and patch urethroplasty with bladder mucosa for a urethral stricture in a hypospadias cripple who has done well. We reconstructed the glans according to the technique of Horton and Devine. However, we had 2 strictures of the urethral meatus that subsequently resolved, 1 stricture of the proximal anastomosis that resolved with internal optical urethrotomy and a fistula in these 8 cases. We approach all patients with stricture disease via the perineal route and try to determine the length of the stricture on the basis of inserting the sound proximally.

Dr. Martin A. Koyle. We try to cut the neourethra flush with the meatus, although we do allow for growth. We measure our graft with the bladder distended and somewhat stretched or at least 10 per cent longer. We then cut off the excess material.

Mr. Philip G. Ransley. I do not believe that we have been right about selecting the correct size because of so many fistulas, although an advantage of this was that we had to re-explore the urethra. A remarkable finding was that the urethra retains its elasticity and balloons if any stenosis of the meatus is encountered. Therefore, it may be that one should actually make the bladder mucosa just a bit smaller rather than larger. I do not think that it contracts in the same way as other free graft material. A trick for the tip of the penis is to excise the end of the indigenous urethra and transpose that to the head of the penis instead of free grafted penile skin. In cases of exstrophy if you are going to perform a single stage repair with bladder neck reconstruction and epispadias repair, it is an opportune time to excise some bladder mucosa for the urethra. As for a pedicle flap, I believe that you could use bladder mucosa as a primary mode of repair and then a pedicle flap at the end. It will be interesting to see how the tip of the original urethra actually works in the case in which it was used.

TABLE 1. *Data base—patients with underlying anomalies*

Age at Presentation	Surgical Exploration	Anatomical Anomaly	Voiding Cystourethrogram	IVP	Ultrasound Studies	Urine Culture	Treatment	Followup (yrs.)
Birth	No	Bilat. vasal ectopia to bladder	Pos.	Neg.	Not done or unobtainable	Pos., organism unknown	Lt.-to-rt. transvasovasostomy, vasal-bladder reimplant	1 episode of epididymitis treated with suppression (8)
6 wks.	Yes	Lt. ureter ectopic to vas, duplicated urethra, rectourethral fistula	Pos.	Pos.	Not done or unobtainable	Pseudomonas	Lt. nephrectomy	No further episodes (15)
8 mos.	Yes	Large prostatic utricle with both vasa entering utricle	Pos.	Pos.	Pos.	E. coli, Enterococcus	Excision of utricle	No further episodes (2)
20 mos.	Yes	Rectourethral fistula	Not done or unobtainable	Not done or unobtainable	Not done or unobtainable	Not done or unobtainable	Anoplasty	1 episode of epididymitis (10)
4 yrs.	No	Urethral-ejaculatory duct reflux	Pos.	Not done or unobtainable	Not done or unobtainable	Pos., organism unknown	Trimethoprim-sulfamethoxazole suppression	Recurrent epididymitis (2)
4 yrs.	Yes	Duplicated urethra with severe stenosis of penile urethra	Pos.	Neg.	Not done or unobtainable	E. coli	Urethroplasty	No further episodes (11)
7 yrs.	Yes	Rt. ureter ectopic to seminal vesicle	Not done or unobtainable	Pos.	Not done or unobtainable	Not done or unobtainable	Rt. nephroureterectomy	No further episodes (7)
8 yrs.	No	Stricture of bulbous urethra with urethral-seminal vesicle reflux	Pos.	Pos.	Not done or unobtainable	E. coli	Ileal conduit urinary diversion, lt. vasectomy	No further episodes (10)

Of the 17 prepubertal patients 8 (47 per cent, or 17 per cent of the total 47 patients), including 3 of 4 infants (75 per cent), had an underlying urogenital anomaly. None of the 26 postpubertal patients had such an anomaly. The younger the patient in this series the greater the likelihood for such an anomaly to be discovered (see figure). All patients with underlying anomalies presented by the time they were 8 years old. Of the 8 patients with an underlying anomaly 5 (63 per cent) underwent scrotal exploration and the diagnosis of epididymitis was made by exclusion.

Data on the patient population with underlying anomalies are reviewed in table 1. The anatomical anomalies included vasal ectopia to the bladder, ureteral ectopia to the vas, ureteral ectopia to the seminal vesicle, a duplicated urethra, bulbous urethral stricture, prostatic utricle, rectourethral fistula and a patient with urethral-ejaculatory duct reflux. One patient had 3 anomalies, including a ureter ectopic to the vas, duplicated urethra and rectourethral fistula. A common theme of these conditions was the entry of infected or sterile urine into the genital ductal system.

The value of clinical presentation, and the presence or absence of fever, leukocytosis, pyuria and bacteriuria in the prediction of patients with an underlying anomaly was examined. Clinical presentation of all patients in the series was remarkably similar and consisted typically of scrotal swelling, erythema and pain. Thus, clinical presentation was of no predictive value. Table 2 details the factors of fever, leukocytosis, pyuria and a positive urine culture, as well as the sensitivity, specificity, and positive and negative predictive values of these parameters in achieving the diagnosis of an underlying anomaly. The presence of fever, leukocytosis and pyuria was of minimal positive predictive value. A positive urine culture had the highest sensitivity (1.00) and specificity (0.79). Every patient with an underlying anomaly in whom a urine culture was available for review (6 of 8) had a positive urine culture. Coliform urinary infections predominated. A positive urine culture has potential in the prediction of those patients with an underlying anomaly (positive predictive value 0.60). A negative urine culture has great potential in predicting patients without an underlying anomaly (negative predictive value 1.00).

Imaging studies were reviewed in an attempt to evaluate the merit of each modality in achieving a diagnostic endpoint (table 3). Of the 47 patients over-all 12 underwent voiding cystourethrography, 12 underwent excretory urography (IVP) and 5 underwent ultrasonography. Of the 8 patients with underlying anomalies 6 underwent voiding cystourethrography and 6 underwent an IVP, and an anomaly was diagnosed in 6 (100 per cent) and 4 (67 per cent), respectively. In 1 patient ultrasonography confirmed the diagnosis of a large prostatic utricle.

TABLE 2. *Predictive value of clinical and laboratory factors*

Factor	No. Pts. Studied	No. Pos.	Sensitivity No./Total (%)	Specificity No./Total (%)	Predictive Value	
					Pos.	Neg.
Fever	47	12	2/8 (25)	29/39 (74)	0.17	0.78
Leukocytosis	13	8	2/5 (40)	2/8 (25)	0.25	0.40
Pyuria	16	9	2/3 (67)	6/13 (46)	0.22	0.86
Pos. urine culture	25	10	6/6 (100)	15/19 (79)	0.60	1.00

TABLE 3. *Efficacy of imaging studies*

Imaging Study	No. Performed		No. Pos./Total (%)
	All Pts.	Pts. With Underlying Anomalies	
Voiding cystourethrogram	12	6/8	6/6 (100)
IVP	12	6/8	4/6 (67)
Ultrasonography	5	1/8	1/1 (100)

In no patient in whom voiding cystourethrography and an IVP were performed was the IVP diagnostic when voiding cystourethrography demonstrated no underlying anomaly. However, 2 patients with nondiagnostic IVPs had diagnostic voiding cystourethrograms. Thus, voiding cystourethrography has the greatest diagnostic yield.

Since voiding cystourethrography best evaluates the lower urinary tract its diagnostic value will be greater in patients with lower urinary tract processes, such as a prostatic utricle, urethral-ejaculatory duct reflux, duplicated urethra, urethral stricture, rectourethral fistula and vasal ectopia to the bladder. The IVP best evaluates the upper urinary tract and its greatest diagnostic value will be in patients with upper urinary tract processes, such as ureteral ectopia to the vas or seminal vesicle. These imaging studies can serve complementary roles, for example voiding cystourethrography demonstrated urethral-ejaculatory duct reflux and an IVP demonstrated bilateral hydroureteronephrosis in the patient with a congenital bulbar urethral stricture. Thus, in the evaluation of epididymitis voiding cystourethrography and an IVP are necessary for lower and upper tract imaging, respectively.

Therapy of patients with underlying anatomical anomalies usually is operative, and involves severance of the pathological connection between the urinary and genital tracts (table 1). Of the 8 patients with an underlying anomaly 7 underwent an operation with satisfactory clinical results. Followup ranged from 2 to 15 years (table 1). Of these 7 patients 5 have had no recurrent epididymitis, while 2 had 1 episode of epididymitis. The patient who was treated nonoperatively has had multiple episodes of recurrent epididymitis and is being considered for unilateral ligation of the vas deferens.

DISCUSSION

Epididymitis is a rare but important differential diagnosis in the infant or child with acute scrotal swelling.[1-4] Our results confirm the rarity of this diagnosis with less than 5 cases per year at a major pediatric center. The diagnosis often is made by findings at scrotal exploration (28 per cent of our patients). As reported by Gierup and associates,[5] our data demonstrate the predominance of coliform urinary tract infections in prepubertal boys with epididymitis. In sexually active male patients from adolescence to an age of 35 years a chlamydial type of organism usually is responsible.[3,6,7]

Although some patients with epididymitis have no demonstrable genitourinary anomaly, our finding of a 35 per cent incidence of anomalies in prepubertal boys suggests that anomalies certainly are not rare.[8] Systemic infection with hematogenous dissemination to the epididymis has been described.[6-9] Hemophilus influenza type B has been implicated,[10] as well as viral,[1,6,8] gonorrheal, tuberculous and nonspecific infections.[8] Post-traumatic[11] and post-instrumentation[1] sources of epididymitis have been described. Retrograde lymphatic spread of infection from the bladder, prostate or posterior urethra to the epididymis has been reported.[6,8] The anti-arrhythmic drug, amiodarone, recently has been associated with the occurrence of epididymitis.[2] When no etiological source for the infection is discovered the term idiopathic epididymitis is applied.

The phenomenon of urethral-ejaculatory duct reflux (retrograde flow of urine from the posterior urethra to the ejaculatory ducts, and then to the vas and/or seminal vesicles) has been implicated in the etiology of acute epididymitis in children.[1,3,7,8,10-12] This condition has been described mainly in children who have undergone instrumentation, or those with chronic infection or bladder outlet obstruction. Reported examples are in association with posterior urethral valves, urethral strictures, posterior urethral masses, duplicated urethra with obstruction, bladder neck obstruction, neurogenic bladder and imperforate anus.[10] Normally, the oblique course of the ejaculatory ducts as they enter the posterior urethra prevents reflux but with severe attenuation of prostatic tissue, as seen with congenital obstruction, urethral-ejaculatory duct reflux can occur.[12] An inflamed or traumatized verumontanum may cause obliteration of the valvular mechanism and permit bacterial entry into the vas.[7] Two of our patients had documented urethral-ejaculatory duct reflux: 1 showed some clinical improvement with oral antibiotics and prophylaxis but he had recurrent episodes of epididymitis and 1 ultimately required ligation of the vas for cure.

Albeit rare, other underlying anatomical abnormalities have been cited as the etiological basis for epididymitis. Ectopic ejaculatory ducts,[7] ectopic ureters emptying into the ejaculatory duct, seminal vesicle or vas deferens,[1,4,13] and a prostatic utricle that communicates with the vas[14] have been described in the literature. Umeyama and associates reported on 5 patients with ectopic ureters who presented initially with epididymitis.[4] Williams and Sago suggested strong consideration of the diagnosis of an ectopic ureter emptying into the genital apparatus when a prepubertal boy with a solitary kidney presented with epididymitis.[13] Ectopic ureters to the genital ducts almost invariably are associated with a dysplastic and nonvisualizing kidney.[14] In a series of 20 boys with prostatic utricles Schuhrke and Kaplan reported that 25 per cent presented with epididymitis.[15] The vas often ends in the wall of the utricle. The presence of a colovesical fistula in association with epididymitis in a child has been described.[7,10]

In our series the common denominator with respect to the underlying anomaly was a pathological connection between the urinary tract and the genital or intestinal tract. This connection provides a means whereby infected or sterile urine may gain access to the male genital ductal system and, therefore, to the epididymis. Our patient who presented in the neonatal period had bilateral vasal ectopia to the bladder that required a transvasovasostomy and nonrefluxing reimplantation of the vas into the bladder. It remains to be seen if this repair will produce viable sperm. Two boys had ureteral ectopia into the genital tract: 1 to the vas and 1 to the seminal vesicle. Both boys had nonfunctioning kidneys and, therefore, nephrectomy was appropriate. One boy had a large prostatic utricle with each vas entering it. Treatment was excision of the utricle and bilateral ligation of the vasa. Two patients had a duplicated urethra, including 1 with severe stenosis of the penile urethra that required urethroplasty. One boy had a bulbar urethral stricture with urethral-ejaculatory duct reflux. Presumably, the distal obstruction led to stasis and reflux to the genital duct system. One patient with an imperforate anus had a fistula between the rectum and posterior urethra, presumably creating a favorable milieu for the development of epididymitis.

It has been well documented that a thorough radiographic evaluation may be of value in children with epididymitis.[1-4,7,16] The key study generally has been the IVP. There is little mention of voiding cystourethrography. Our series clearly demonstrates that voiding cystourethrography is an excellent means to diagnose the underlying anomaly. Ultrasonography may well prove to be a reasonable means of evaluation of the upper tracts if voiding cystourethrography is negative.

In conclusion, epididymitis is a rare entity in children, the diagnosis of which often is made at scrotal exploration. Underlying anatomical genitourinary anomalies are not rare, being present in 75 per cent of the infant population and 47 per cent of the prepubertal population presenting with epididymitis. Particularly, if the urine culture is positive an underlying anomaly is likely to be discovered.

We advocate that any prepubertal patient presenting with epididymitis, especially those with a positive urine culture, merits a complete urological evaluation, including voiding cystourethrography and an IVP. Indeed, 7 of our 8 patients (88 per cent) with an underlying anomaly responded to an operation with favorable long-term clinical results.

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