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Testicular Torsion in Undescended Testis : A Case Report and View of Sixty-two Cases in Japan

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ABSTRACT : A 7-year-old boy with cerebral palsy was presented with a progressively enlarging tender, left inguinal mass. Examination revealed absence of the left testis in the scrotal sac. The left testis was palpable in the left superficial inguinal pouch. The right testis was normaly palpable in the right scrotum. A diagnosis of a torsion in the undescended left testis was confirmed by exploratory surgery. Orchidectomy of the left testis was performed and the remainder of the patient's course was uneventful. We presented this case to describe the clinical features of testicular torsion in the undescended testis with a review of 62 cases in Japan. Specific emphasis was placed on the incidence, relationship of cerebral palsy to torsion, diagnosis, and treatment of testicular torsion in cryptorchidism.

INTRODUCTION

Testicular torsion in the undescended testis is a very rare disease. Most series dealing with the undescended testis (cryptorchidism) have reported the incidences of testicular torsion at 7.9-13.3% in Europe and the United States[1~4] and 7.1-9.1% in Japan[5~8]. A high incidence of cryptorchidism has been also noted in patients with cerebral palsy[3, 4]. In the present paper, a case of testicular torsion in the undescended testis with cerebral palsy was reported with a review of the clinical features of 62 cases with torsion of the testis in cryptorchidism which were cummulated in Japan.

CASE REPORT

A 7-year-old boy was admitted to our hospital in January 23, 1991 with a three-day history of a progressively enlarging tender mass in the left inguinal region. The symptoms were associated with poor nutritional intake, occasional nausea, vomiting and increased temparature. The patient had profound mental and physical developmental retardation and a history of seizure. He had a perinatal history that the umbilical cord twined around his neck on birth, which was thought to have resulted in his cerebral palsy. He had been seen several years prior to admission by his family physician, who thought that the left inguinal mass represented an undescended testis.

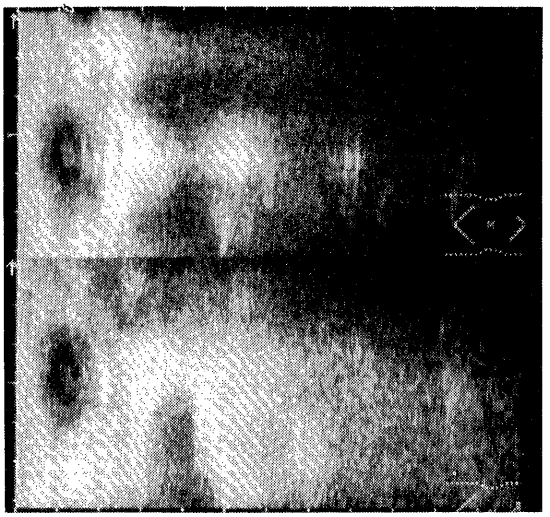

Fig. 1. Ultrasonography

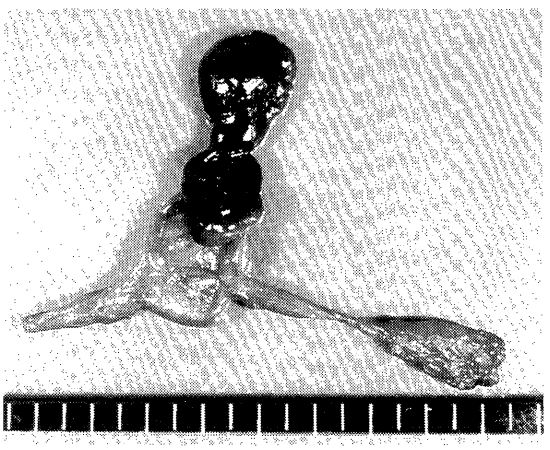

Fig. 2. Resective specimen

On physical examination the patient was a thin, poorly developed boy who appeared to be appromixately 4 years old and weighed 17.5kg. He reacted to painful stimuli, but his vocalizations were unintelligible. Vital signs were as follows : blood pressure, 100/60 mmHg ; pulse, 98/min ; respirations, 22/min ; and temperature, 37.5 C.

Significant findings aside from the patient's mental retardation included the following: a soft, nontender abdomen with active bowel sounds and no masses or hepatosplenomegaly; spastic quadriplegia with flexion contractures of all four extremities without hip dislocations but with adduction contractures and scissoring of the legs ; and a tender mass measuring approximately 2cm in diameter in the left inguinal region. The right testis was normaly palpable in the right scrotum.

White blood cell count was 16,100 and the remaining laboratory data (red blood cell count, electrolytes, BUN, blood glucose, urinalysis) were within normal limits. Ultrasonography on the left inguinal mass showed a hypoechoic mass with some strong central echogenicity, which suggested the torsion of the undescended testis (**Fig. 1**).

The operation was performed on January 28, 1991. Exploration of the left groin revealed a 180 counter-clockwise intravaginal torsion of an undescended testis in the left inguinal pouch. The testis was found to be necrotic and an orchidectomy was performed (**Fig. 2**). During anesthesia it was noted that the right testis descended into the scrotal sac. Histologic examination confirmed that the mass was a necrotic torsioned testicle. The patient had an uneventful postoperative course and was discharged on the tenth hospital day.

DISCUSSION

Testicular torsion was first reported by Delasiauve in the United Kingdom in 1840[3]. His initial patient was a 15-year-old boy with torsion of an undescended testis. In 1904 Yamamura reported the first case in Japan affecting an undescended testis[8], and maldescent has since been recognized as increasing the risk of torsion[1~6]. Williamson suggested that torsion was approximately ten times more common in cryptorchidism[9]. The most common factors predisposing to the testicular torsion are congenital anatomical anomalies of the testis, and the undescended testis, which is accompanied with these abnormalities, are more prone to torsion[9, 10]. In up to 7% of male infants maldescent occurs and the testis come to rest in the inguinal canal (70%) or in the extraperitoneal tissues or abdomen (25%), and the remaining 5% descend into various other areas, that is called the ectopic testis[9~11]. In the present paper 62 cases with testicular torsion in the undescended testis in Japanese publicatiions up to 1990 were

accumulated with sequal to the previous series reported from Watanabe *et al.*[12~17].

The frequency of testicular torsion was higher in the teen-age group than in infants below 10 years of age, whereas the torsion of the undescended testis showed a similar incidence in both age groups (**Table 1**). Delay of diagnosis is common, markedly reducing the salvage rate of the affected testis. Only 21 cases (33.9%) were diagnosed acculately on addmission, and the others were missed such as strangulated inguinal hernia, acute appendicitis, orchitis and so on (**Table 2**). Clinical symptoms contributing to testicular torsion of the undescended testis have been pointed out[16]: 1) sudden onset of lower quadrant pain, with poor spirit, anorexia and vomiting in some infants below 2 years of age, 2) presence of a hot, red, and progressively enlarging tender mass in the inguinal canal or at the external inguinal ring, 3) absence of intrascrotal testis, 4) localized pain at the mass without peritoneal irritation, 5) good condition with a low-grade or no fever. The differential diagnosis between torsion and other causes for the acute inguinal mass could be achieved by means of ultrasonography (US) or radionuclide imaging[18, 19]. US could evaluate pathological features of the testis and its location. Testicular torsion of the undescended testis were hypoechoic with some strong central echogenecity, reflecting the pathological changes such as parenchymal bleeding or necrosis. The diagnosis of torsion of an undescended testis should be considered in any patient with above symptoms and signs, using the help of imaging modalities.

On the other hand, testicular maldescent may be associated with such entities as genetic syndromes, intersex, hormonal defects, renal anomalies, mechanical gubernacular defects, inguinal hernias, Wilms's tumor, torsion, and vasal and epididymal abnormalities[11, 20]. Ankerhold *et al* noted a high incidence (43.7%) of cryptorchidism in 87 patients with cerebral palsy[4]. Cerebral palsy has been thought to occur as a result of birth trauma, neonatal asphyxia, or postnatal problems, and damage to the fetal hypothalamo-pituitary axis during complicated pregnancies may result in testicular maldescent[3, 4, 9~11]. Our case had a perinatal history of twined umbilical cord around his neck at birth, but there does not seem to be not only a relationshpip between cremasteric muscle spasm and maldescent, but also a relationship of torsion infarction in undescended testis to spastic neuromuscular disease[3]. Our case illustrates the need to include torsion of an undescended testis in the differential diagnosis of an inguinal mass, especially in those patients with cerebral palsy. Familiarity with this lesion will help to differentiate it from other causes of inguinal mass, which was suspected initially in our patient.

Damage to the gonad varies with the degree of torsion, its tightness, and its duration. It has been demonstrated experimentally in dogs that four complete twists (1440 degrees) of the spermatic cord produce irreversible changes within 2 hours, whereas one turn (360 degress) produces no changes up to 12 hours[21]. In Japanese series, there were no significant differences among the direction of the testicular rotation, that was clockwise rotation in 21 and counterclockwise rotation in 19 cases. The degree

Table 1. Age Distribution of Testicular Torsion in Undescended Testis

Age (years)	No. of Cases
0– 5	10
6–10	13
11–15	9
16–20	13
21–25	5
26–30	2
31–35	2
36–40	2
41–45	4
46–50	2
Total	62

Table 2. Preoperative Diagnosis of Testicular Torsion in Undescended Testis

Preoperative Diagnosis	No. of Cases (%)
Testicular torsion	21 (33.9)
Strangulated inguinal hernia	8 (12.9)
Appendicitis	2 (3.5)
Orchitis	1 (1.7)
Undescended testis	1 (1.7)
Unknown	29 (46.7)
Total	62 (100%)

Table 3. Degrees of Testicular Torsion in Undescended Testis

Degrees of Torsion	No. of Cases
180	15
200	1
270	3
310	1
360	20
540	2
720	5
900	3
Total	50

Table 4. Treatment of Testicular Torsion in Undescended Testis

Treatment	No. of Cases
Orchidectomy	51
Detorsion	7
Spontaneous detorsion	1
Conservative	1
Total	60

Table 5. Relationship of Operative Procedure to Duration Times from Onset (No. of Cases)

Duration Times (days)	Operative Procedures		Total
	Orchidectomy	Detorsion	
-1	3	3	6
1-2	10	1	11
2-3	8	1	9
3-4	0	1	1
4-5	3	1	4
over 5 days	11	0	11

of rotation ranged from 180 degree to 900 degree, with the high frequencies of 360 and 180 degree of rotation (**Table 3**).

Immediate surgical exploration is the treatment of choice to reduce torsion of an undescended testis. Fifty-one out of 62 cases (83.5%) in Japanese series underwent orchidectomy for necrotic testis with complete vascular occlusion (**Table 4**). However, there were a few variabilities with some patients having operative and spontaneous detorsion within 5 days (**Table 5**). Wiliamson's experience revealed that torsion of the undescended testis occurred most commonly in the superficial inguinal pouch, followed by incidences anatomically at the site of the external inguinal ring[9]. Orchiopexy should be performed if the involved testis is viable, and prophylactic fixation to the scrotum of the contralateral testis is recommended.

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