CASE REPORT

Splenogonadal fusion misdiagnosed as spermatic cord cyst: a case report

Fusion splénogonadique prise pour un kyste du cordon spermatique: Cas clinique

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Abstract

Introduction : Splenogonadal fusion is a rare congenital anomaly. The aim of this study was to report a case of splenogonadal fusion mimicking a spermatic cord cyst, and discuss therapeutic management of this rare congenital malformation.

Observation : An eight-years old patient was presented with an asymptomatic three-centimeter oval scrotal mass mistaken for a spermatic cord cyst. Surgical exploration has revealed tow purple-red, firm encapsulated masses. The first mass was two cm long and adherent to the upper pole of the left testis with a cleavage plane. The second mass was four cm long, attached to the first by a fibrous cord and drawn on its superior pole by a serpiginous vascular structure that extended inside the abdomen. The spermatic cord was individualized. Extemporaneous anatomopathological examination of the first mass, totally excised, has concluded to benign lesion. Therefore, the peritoneum was opened, and the superior mass was excised as high as it could be reached without orchiectomy. Definitive Anatomopathological examination concluded to an ectopic splenic tissue. The final diagnosis was a continuous splenogonadal fusion.

Conclusion : This case highlights the clinical characteristics of this condition, with a special focus on the signs and findings that might help prevent unnecessary orchiectomy. Consequently, it is essential to include this malformation in the differential diagnosis of scrotal masses in children.

Key words: Cryptorchidism; Spleen; Testis.

Résumé

Introduction : La fusion splénogonadique est une anomalie congénitale rare. Le but de cette observation était de rapporter un cas de fusion splénogonadique imitant un kyste du cordon spermatique et de discuter la prise en charge thérapeutique de cette rare malformation congénitale.

Observation : Un patient de huit ans a consulté pour une masse inguinale gauche ovale asymptomatique de trois centimètres qui a été prise pour un kyste du cordon spermatique. L'exploration chirurgicale avait révélé deux masses encapsulées fermes et rouge pourpre. La première masse mesurait deux cm de long et adhérait au pôle supérieur du testicule gauche avec un plan de clivage. La deuxième masse mesurait quatre cm de long, attachée à la première masse par un cordon fibreux et se continuant sur son pôle supérieur par une structure vasculaire serpigineuse qui s'étendait en intra-péritonéale. Le cordon spermatique a été individualisé. L'examen anatomopathologique extemporané de la première masse, totalement excisée, concluait à une lésion bénigne. Par conséquent, le péritoine a été ouvert et la masse supérieure a été excisée aussi haut qu'elle pouvait être atteinte sans orchidectomie. L'examen anatomopathologique définitif avait conclu à un tissu splénique ectopique.

Conclusion : Ce cas clinique souligne les caractéristiques cliniques de cette malformation, et met l'accent sur les spécificités qui pourraient aider à prévenir une orchidectomie inutile. Par conséquent, il est essentiel d'inclure cette malformation dans le diagnostic différentiel des masses scrotales chez l'enfant.

Mots clés : Cryptorchidie; Rate; Testicules.

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INTRODUCTION

Splenogonadal fusion is a rare congenital anomaly (1). Reported in less than 200 cases in literature, several theories postulate that it would occur between the 4th and the 5th week of gestation (1, 2). More reported in male, it is usually diagnosed incidentally during exploration of a cryptorchidism or surgery of a hernia (1, 2). Diagnosed wrongly as a para-testicular tumor, it may, sometimes, lead to an inadvertent orchiectomy (1, 2).

We, herein, report a case of splenogonadal fusion mimicking a spermatic cord cyst.

OBSERVATION

The present case was reported according to the CARE Guideline (3).

We report the case of an eight-year old boy, referred to our pediatric surgery department for left spermatic cord cyst. An informed consent was received from the parents.

Physical examination showed a three cm oval swelling on the left inguinal region. This swelling was painless and not reducible. The light transillumination test was not done. Left testicle was in his normal location in the scrotum.

Physical examination did not reveal any other abnormality. The patient was operated under general anesthesia, by an inguinal approach. No investigation was performed.

Instead of the spermatic cord, the surgery revealed two purple-red, firm encapsulated masses. (Figure 1) A first mass of two cm long, adherent to the upper pole of the testicle but separated from it by a cleavage plane. Then, the second mass was four cm long, fusiform, attached to the first mass by a fibrous cord, its superior pole extended inside the peritoneum. The left testicle, located in the scrotum had a normal size and aspect. The spermatic cord was well identified and individualized.

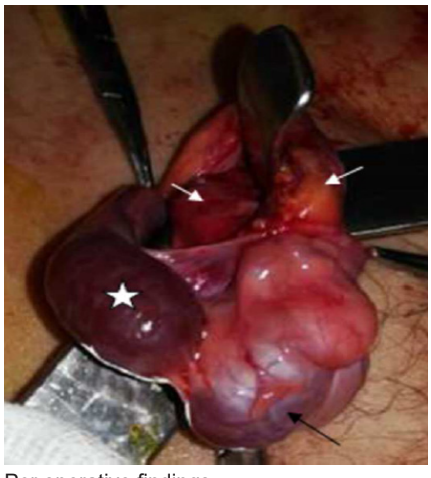

Figure 1. Per operative findings:
☆ : Well-limited cleavable mass for extemporaneous examination.
Left white arrow: Second mass extended inside the peritoneum
Right white arrow: Spermatic cord
Black arrow: Testicle

The presence of the second mass with its extension inside the peritoneum and the presence of a well individualized cleavage plan, suggested benign nature, motivating, as a first step, the opening of the peritoneum and the excision of the superior mass as high as it could be reached.

Extemporaneous examination of the specimen revealed as expected, a benign tissue without specifying its nature. Therefore, we excised the second mass without orchidectomy and performed a herniotomy.

Postoperative abdominal ultrasonography was normal. Definitive pathology concluded to an ectopic splenic tissue and the excised specimen was free from testicular tissue. The final diagnosis was a continuous type of splenogonadal fusion.

A two-year post-operative follow up showed a left testicle in its scrotum. No difference in size in comparison to the contralateral testicle was noticed.

DISCUSSION

Splenogonadal fusion is a rare congenital anomaly (1, 2). It is most often diagnosed during exploration for cryptorchidism or hernia repair. Usually it occurs on the left side and is often associated to ipsilateral cryptorchidism (1, 2). In our case, it mimicked a spermatic cord cyst and did not prevent the testicle from continuing its normal migration into the scrotum.

The diagnosis could have been suspected if we had done the light transillumination test. The negativity of the light transillumination test would have prompted us to further investigate and request an ultrasound. The fact that unnecessary orchiectomy was still performed in 21.95 % (2) of splenogonadal fusion patients shows the ongoing need for better diagnostic procedures and increased public knowledge of this illness. Clinically, an inguinal-scrotal lump that has been present since birth and has been growing slowly and benignly for years should be evaluated for splenogonadal fusion. Radiologically, preoperative ultrasound would have been a great contribution to determine the non-cystic echogenicity of the swelling. Usually, it shows a paratesticular mass, difficult to be differentiated from neoplastic masses, but sometimes, it could, in case of continuous form, visualize the cord linking the spleen to the testicle (4, 5). The scintigraphy using technetium-99m, when it is available, could make the diagnosis with certainty (4, 5).

Two types of splenogonadal fusion are described in the literature. In our case, the fusion was continuous, which is described to be the most common type and must evoke the diagnosis (6, 7). Discontinuous splenogonadal fusion may rather lead to the diagnosis of testicular tumor and pushes towards the practice of orchiectomy (6, 7). Many theories have attempted to explain this malformation (1, 9). While the presence of discontinuous forms and rare cases of right splenogonadal fusion could argue in favor of a migrating accessory spleen (8, 9), the size of the splenic masses in certain splenogonadal fusions, which can sometimes exceed the average reported in the literature, of one to two

centimeters, would rather plead in favor of the inflammatory theory (8, 9). In most cases of reported splenogonadal fusion, there was a single ectopic spleen (9). In this case, the presence of two masses separated by a fibrous cord made an extemporaneous anatomopathological examination possible

The decision whether or not to perform a complete excision of the splenic tissue is difficult, as hyperplasia of the splenic tissue left in place can cause testicular atrophy and on the other side, an excision can compromise testicular vascularization (2, 8, 9). Our present case of splenogonadal fusion in a boy misdiagnosed as a spermatic cord was not unusual for known splenogonadal fusion cases. Lack of awareness of this condition is a major reason for its misdiagnosis. What made our case unusual was the presence of two masses. Splenic tissue was separated from the testicle by a cleavage plane, and the spermatic cord was easily distinguished. The excision was easy and did not damage neither the testicular tissue nor the spermatic cord. The follow up showed a normal size of the testicle.

CONCLUSION

As splenogonadal fusion is a benign tumor, we recommend the least damaging treatment for the testicle. Extemporaneous examination when it's possible helps in decision making.

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