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该研究依托上海交通大学医学院“双一流”暨高水平地方高校建设“一流学科—临床医学－临床研究中心建设”项目。

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The research relies on the Project of Clinical Research Center, Clinical Medicine, First-Class Discipline of "National Double First-Class" and "Shanghai Top-Level" high education initiative at Shanghai Jiao Tong University School of Medicine.

病例报告

连续型脾性腺融合 1 例报道及文献复习

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[摘要] 患儿，男，4 岁，因左侧阴囊出现无痛性团块 3 年收治入院。查体左侧腹股沟至阴囊依次可及 3 个类似睾丸样团块，上方及中部质地稍硬，下方质地稍软。入院后腹腔镜探查发现左侧有脾性腺融合（splenogonadal fusion，SGF），行保留左侧睾丸副脾切除术。术后随访 18 个月，睾丸发育良好、无萎缩，腹部 B 超提示肝、胆、脾未见异常。SGF 是少见的先天异常，术前诊断困难，术中发现 SGF 应采取保留睾丸副脾切除术。腹腔镜技术可作为一个有效的诊治方法，其可提高对 SGF 的认识，避免不必要的睾丸切除术。

[关键词] 脾性腺融合；儿童；腹腔镜；保留睾丸副脾切除术

[DOI] 10.3969/j.issn.1674-8115.2020.07.023　[中图分类号] R726.9；R697+.22　[文献标志码] B

Continuous splenogonadal fusion: a case report and review of literature

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[Abstract] A 4-year-old boy with a painless mass for 3 years in the left scrotum was admitted to the hospital. Three testicle-like substances could be palpable one by one from groin to scrotum on the left side during physical examination. The upper and middle parts were slightly hard, while the lower part was slightly soft. The splenogonadal fusion (SGF) in the left side was found by laparoscopy after admission. Accessory splenectomy was performed with preservation of the left testicle. Postoperative follow-up was 18 months. The testicles were well developed without atrophy, and abdominal B-ultrasound showed no abnormality in liver, gallbladder and spleen. SGF was a rare congenital abnormality, which was difficult to diagnose preoperatively. During the operation, if the SGF was found, accessory splenectomy with testicle-sparing should be performed. Laparoscope can be used as an effective diagnosis and treatment method to improve the understanding of SGF, and avoid unnecessary orchiectomy.

[Key words] splenogonadal fusion (SGF); children; laparoscopy; accessory splenectomy with testicle-sparing

脾性腺融合（splenogonadal fusion，SGF）是临床少见的先天性畸形，主要表现为脾脏组织与生殖腺或中肾管残迹的异常融合[1]。因其病例少见，术前影像学检查较难做出明确诊断，临床上可能出现误诊以及不必要的睾丸切除[2]。现结合文献将上海市儿童医院 / 上海交通大学附属儿童医院泌尿外科诊治的 1 例 SGF 病例报告如下。

1　临床资料

1.1　临床表现及查体

患儿，男，4 岁，出生后 1 年发现左侧阴囊无痛性团块，未予处理。后因团块 3 年未缩小，于我科进一步诊疗。查体：心肺正常，腹平软；左侧腹股沟至阴囊依次可及 3 个类似睾丸样团块，其上方及中部质地稍硬，下方质地稍软，位于阴囊中上部，周边有囊性感，无压痛，不可还纳。术前诊断左侧多睾症可能，并左侧鞘膜积液。

1.2　实验室及影像学检查

肿瘤标志物、内分泌检查未见明显异常。腹部 B 超检查提示肝、胆、脾未见异常；腹腔、腹股沟淋巴结未见明显异常；左侧腹股沟至阴囊内自上而下见 3 个低回声团，大小分别为 10 mm × 4 mm × 6 mm、15 mm × 7 mm ×

[基金项目] 上海市市级医院新兴前沿技术联合攻关项目（SHDC12010108）。

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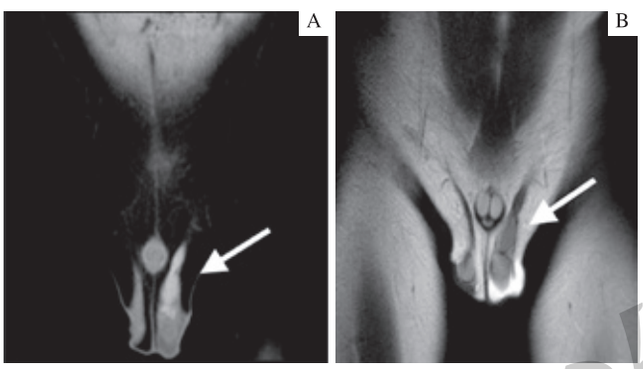
8 mm、15 mm × 7 mm × 8 mm，左侧阴囊内探及无回声区。B 超提示左侧多睾可能，左侧鞘膜积液。盆腔 CT 增强扫描提示左侧睾丸上方占位，左侧鞘膜积液，左侧精索增粗（图 1A）。盆腔磁共振成像（magnetic resonance imaging，MRI）增强扫描提示左侧阴囊 2 枚睾丸信号，上方 1 枚异常信号（图 1B）。



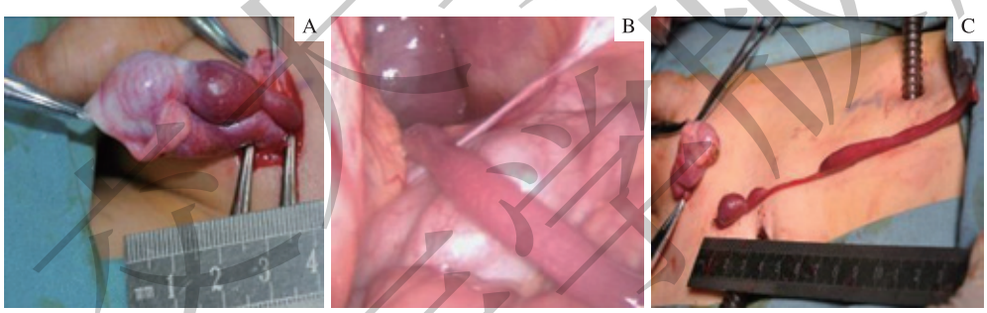
Note: A. Coronal CT enhancement. B. T2 weighted MRI.

图 1 盆腔 CT 增强及 MRI 增强扫描图像

Fig 1 Enhanced CT and enhanced MRI images of pelvic cavity

1.3 治疗和随访

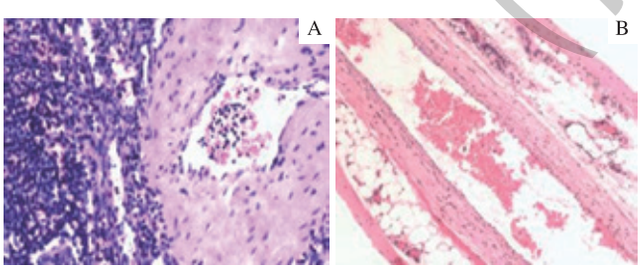
患儿完善术前检查行腹腔镜探查，见左侧条索状棕红色组织，近端发自脾门，远端变细并通过孔状未闭合鞘状突至腹股沟；腹股沟探查见棕红色条索增粗，远端连接棕红色团块并与睾丸上极相连，团块与睾丸边界清晰，睾丸颜色正常，考虑棕红色团块为副脾。术中所见增粗条索、团块，睾丸与术前超声描述基本相同，近端 2 枚质地稍硬，远端（睾丸）质地稍软。腹腔镜下近端高位双重结扎条索并切断，从腹股沟切口将条索牵出切口外。沿团块与睾丸连接部切开包膜表面，将副脾从睾丸上极游离以保留完整睾丸组织，松解后左睾丸可位于阴囊底部，同时行左睾丸下降固定术、左侧鞘状突高位结扎术（图 2）。术后苏木精 - 伊红染色（hematoxylin-eosin staining，H-E 染色）石蜡切片证实为副脾（图 3）。手术顺利，术后第 1 日出院。最后确诊为连续型 SGF。术后半个月门诊复查，血常规正常，腹部 B 超检查肝、胆、脾未见异常，睾丸位于阴囊内，无红肿、感染等并发症。术后随访 18 个月，睾丸发育良好、无萎缩，腹部 B 超提示肝、胆、脾未见异常。



Note: A. Continuous-type SGF was observed intraoperatively. B. Splenic cord was observed laparoscally. C. Testicle, accessory spleen and splenic cord were observed postoperatively.

图 2 连续型 SGF 手术中图像

Fig 2 Intra-operative surgical procedures of continuous-type SGF



Note: A. H-E staining of accessory spleen (× 100). B. H-E staining of fibrous band (× 40).

图 3 副脾组织石蜡切片 H-E 染色

Fig 3 H-E staining of SGF tissues

2 讨论

SGF 是临床罕见的先天性畸形，多见于男性，约 97% 的病例发生在左侧 [3]。70% 的病例发病年龄小于 20 岁，50% 的病例小于 10 岁 [4]，男女发病率的比例为 15.0:1 ～ 16.6:1[5-6]。根据异位脾组织和脾脏之间有无索带连接，SGF 分为连续型和不连续型 [7]。早期研究 [4] 显示 2 种类型的 SGF 发生比例相近，但近年来研究 [8] 表明连续型 SGF 比例更高。连续型 SGF 中，有一条索状脾样组织或纤维组织从原位脾的上极向睾丸延伸；不连续型 SGF 中，脾脏和性腺之间不存在异常连接 [4, 9]。本病例是 1 例 4 岁男性左侧连续型 SGF。

SGF 具体发病机制尚不清楚。胚胎发育 5 ～ 8 周，性腺尚未下降时，脾脏和性腺发生融合，同时覆盖于脾脏和生殖脊的腹膜表面的轻度炎症可能导致这 2 个器官部分融合[3]。约 50% 的连续型 SGF 伴有先天发育异常，常见伴发隐睾、四肢不全、小颌畸形，少见合并先天性心脏病、腭裂、肛门闭锁、脊柱裂，而不连续型 SGF 很少合并上述先天发育异常[10]。本病例虽为连续型 SGF，也未见上述相关异常。

SGF 是一种罕见的良性疾病。由于 SGF 可干扰正常的睾丸下降及鞘状突闭合[11-12]，故大多数 SGF 病例是在腹股沟疝、隐睾或睾丸肿块等手术探查中偶然诊断发现[5]。影像学检查主要包括 B 超、CT、MRI 和同位素检查。Karray 等[2]报道 B 超及 MRI 对 SGF 检查不够精确，敏感度及特异度不高，术前很难做出精确诊断；Jakkani 等[8]和 Bosnalı 等[13]认为 CT 及锝 -99m（^{99m}Tc）同位素扫描对 SGF 有一定诊断价值，但相关报道很少。本病例术前同样未确诊，曾考虑多睾症可能，但因体检提示各团块质地不均匀，故采用腹腔镜探查才确诊为 SGF。

目前，对于 SGF 采取保守观察还是手术治疗仍存在争议。有文献[4]指出 SGF 是良性病变，如果不存在临床症状，无需手术治疗。另有文献[2]报道 SGF 虽然为良性病变，但仍需手术探查，并需术中病理组织学检查以排除恶性肿瘤。手术一般采用腹腔镜探查，对连续型 SGF 术中可同时切除索带以避免并发肠梗阻。由于术前缺乏对 SGF 解剖的正确认识，仍然有 37% 的 SGF 患者错误地接受睾丸切除术[14]。实际上脾组织通常可以很容易地从性腺中分离出来[15]，手术应采取保留睾丸的副脾切除术。Srinivasa 等[14]报道副脾两侧纤维索带扭转 360° 情况下，仍可顺利保留睾丸而仅行副脾切除。本例因受异位副脾牵拉，睾丸位置上移，睾丸下降受到影响，切除副脾及索带即可使精索延展，解决睾丸未降问题。

SGF 临床症状不典型，多数没有临床症状，少部分表现阴囊肿大、肿块，个别病例表现为疼痛、睾丸炎、睾丸扭转、异位脾组织外伤性破裂或腹腔内脾索造成的肠梗阻[16]，易误诊、漏诊，故需要进一步提高对 SFG 的认识。结合文献，总结以下经验：①本病例表现阴囊无痛性多发团块，需要排除阴囊肿瘤，可以首选超声及肿瘤标志物进行鉴别。②超声虽然考虑多睾症可能，但查体发现团块质地与睾丸有一定差别，副脾质地比睾丸质地更硬。③本例术前仅做盆腔扫描，脾索在下腹部扫描中止时被误认为增粗的精索；术前如考虑 SFG，增加腹部扫描可以发现脾索与脾脏的关系从而帮助诊断；进而行同位素扫描可进一步确诊。④术前如考虑 SFG，建议进行腹腔镜探查以明确诊断；对于连续型 SFG，腹腔镜下高位结扎脾索操作更为方便；因 SFG 合并内环口未闭，切断脾索后可以很容易在腹股沟切口牵出脾索。⑤保留睾丸手术应作为首选，副脾与睾丸有明显边界，可以很容易将副脾从睾丸上分离。

综上所述，SFG 是一种罕见的先天性畸形，临床诊断困难；腹腔镜可作为一个有效的诊治方法，且还需提高医师对 SFG 的认识，避免不必要的睾丸切除术。

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[收稿日期] 2019-12-24 [本文编辑] 崔黎明