·Case report·

Rectal duplication combined with teratoma: a case report
直肠重复畸形合并畸胎瘤一例报告

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[**KEY WORDS**] rectal duplication; teratoma; sugrery
[**CLC number**] R 574.63; R 730.269 [**Document code**] B [**Article ID**] 0258-879X(2004)10-1159-02

* **Clinical data** An 18-year-old girl presented with difficulty in passing urine and lower abdominal mass for 2 months. Fifteen days before admission to our hospital, she received laparotomy in another hospital during which a mass was found in retroperitoneal space. The doctor ceased further exploration and closed the abdominal wall due to lack of experience, then the girl was sent to our hospital for further treatment. On examination the patient was well built with no physical abnormalities except that the right side of the hip was a little higher than the left side; on rectal digital examination a mass outside the rectum about 5 cm from the anal verge on the right was touched. It was soft and nontender but the examining finger could not go above it. Intravenous pyelography showed that the left ureter was pushed aside and the left superior part of the bladder was depressed. CT scanning revealed a huge mass behind the uterus and the sigmoid colon with no infiltration into the surrounding structures and its texture was like that of a soft tissue. Another mass was found to be inferioposterior and to the right of the first mass, pressing the surrounding tissues. No other special familial history, personal history and inherited diseases were found. About 20 days after the first operation, another laparotomy was performed under general anaesthesia during which a fully encapsulated mass, 7-8 cm in diameter, was found beneath the pelvic floor when the peritoneum over it was opened. During the dissection, the mass, measured about 5-7 cm at its widest diameter, was formed with curved tube-like structure attached to its own messentery, blinded at its superior free end, and inferiorly attached to the junction between rectum and the sigmoid colon. The second mass was about 7 cm in diameter and sited between the rectum and the sacrum, which was attached to the surrounding structures and protruded out through the right hip. Its content was yellowish jelly-like and contained hair-like structures. After careful dissection between the masses and their surrounding structures and ligating the tiny blood vessels, the 2 masses were removed and the abdomen was closed. No complications occurred after operation and the patient was discharged on the tenth post-operational day. Pathological report: the wall thickness of the tube-like structure varied from 1-10 mm and contained nonstraited muscles and intestinal mucosa with the characteristic of gastrointestinal tract. These characteristics concorded with the diagnosis of rectal duplication. The second mass comprised of squamous cells, appendages of skin and striated muscles and fat, thus diagnosed as teratoma.

Discussion Rectal duplication is part of the Notochord syndrome in which various abnormalities like teratoma, rectal duplication, minigocele, *etc* co-exist, accounting for about 3% of all enteric duplications. They are mainly located on the mesenteric border of the bowel and may be separated from the lumen, but the curved rectal duplication is relatively a rare phenomenon. Symptoms varied from abdominal pain to that caused by pressing surrounding structures.

Teratoma arises from embryonic stem cell layers like: ectoderm, mesoderm and the endoderm, and therefore has its constituent elements as the skin and its appendages, sebaceous materials, smooth muscles, bone, tooth, fat and gastrointestinal epithelium. It is a slow growing tumor mostly presenting at birth. They become symptomatic only when they are big enough to cause pressure symptoms. They are mostly found in the sacrococcygeal region.

Benign neoplasm takes time to be symptomatic. At the time when it is large enough to cause pressure symptoms, the patient may be at quite an advanced age though the tumor could have been present at birth. The possibility of a malignancy is rare and the congenital disease should be firstly considered. They are normally insidious in presentation as evident in the patient under discussion. Although there are others examination like barium enema, CT has become the

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most important tool in the diagnosis and management of abdominal surgical conditions. It can give message in evaluating the trait of the mass, defining the surrounding structures and their relationship with the mass, thus being valuable in evaluating the resectabilty of the mass.

Patient with symptoms of pressure indicates a laparotomy. Even the nature of the mass was not known, it is important that the dissection should be continued until there are very important vessels or structures were found involved. In this case, though the mass is very large, because the capsula is intact, the dissection prove to be not too difficult, and the 2 masses are all removed perfectly. The patient recovered well and the symptoms disappeared after the operation and the follow-up indicated that the prognosis was good.

[**Received**] 2004-07-27 [**Accepted**] 2004-09-08
[**Editor**] YU Dang-Hui

· 个案报告 ·

重症肌无力合并多发性神经根神经病一例报告

Myasthenia gravis complicated with polyradiculoneuropathy: a case report

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[关键词] 重症肌无力; 胸腺瘤; 多发性神经根神经病
[中图分类号] R 746.1; R 745.4 [文献标识码] B [文章编号] 0258-879X(2004)10-1160-01

1 临床资料 患者男性, 48岁, 因先后四肢无力伴睁眼及吞咽困难10个月, 四肢麻木6个月, 于2003年9月入院。2002年11月中旬起, 无诱因四肢乏力, 活动后加重, 休息后减轻, 1周后出现睁眼费力, 继而吞咽困难, 咀嚼无力, 进餐时间延长。外院诊断为"重症肌无力", 服溴吡斯的明有效, 双手能举过肩。但胸部MRI检查发现胸腺瘤伴左肺转移, 遂于2003年1月行胸腺瘤及左肺楔形切除术。术后胸部放疗, 共33次。放疗期间四肢肌力好转, 但出现左侧周围性面瘫。放疗结束1周后, 出现四肢麻木, 且肢体无力加重, 服溴吡斯的明不能缓解, 2个月后行肌电图检查发现, 周围神经的运动及感觉传导速度减慢, H反射不能引出, F反应延长, 提示"周围神经及神经根损害(轴索损害)"。肌无力症状仍继续加重, 入院前生活自理困难, 但呼吸不受影响。入院时查体, 左侧轻度周围性面瘫, 四肢远端肌肉轻度萎缩, 肌力5$^-$级, 共济运动正常, 四肢痛温觉及深感觉正常, 腱反射对称减弱。病理征阴性。辅助检查: 三大常规、肝肾功能及血糖正常。血清乙酰胆碱受体抗体5.95 P/N(正常值2.2 P/N), 胸腺瘤相关抗体7.35 P/N(正常值2.2 P/N)。脑脊液检查: 细胞总数4×10^6/L, 有核细胞数为0, 蛋白定性(+)、定量1.26 g/L, 氯化物125 mmol/L, 糖3.1 mmol/L, IgG 101 mg/L, IgA 11.1 mg/L, IgM 1.09 mg/L。肌电图: 低频重复刺激面神经递减21%～24%, 腋神经递减45%～51%; 周围神经运动及感觉传导速度减慢。入院后予维生素B_{12}、B_1营养神经, 表柔比星、长春新碱、环磷酰胺及地塞米松联合化疗1个疗程, 1个月后四肢无力及肢体麻木症状明显好转, 生活能自理, 吞咽正常。再次进行联合化疗, 复查血清乙酰胆碱受体抗体3.89 P/N, 胸腺瘤相关抗体3.34 P/N。目前随访, 四肢肌力正常, 仅残留足趾麻木及左侧轻微周围性面瘫。

2 讨 论 重症肌无力是一种自身免疫性疾病, 主要累及神经肌肉接头突触后膜乙酰胆碱受体。约75%的患者合并胸腺异常, 其中15%合并胸腺瘤。重症肌无力合并其他免疫性疾病, 如系统性红斑狼疮、类风湿性关节炎、甲状腺功能亢进等国内外已有报道, 但合并多发性神经根神经病则极罕见。本例具有典型的重症肌无力表现: 累及全身多数骨骼肌的病态疲劳, 活动后加重, 休息后好转, 症状晨轻晚重, 服用胆碱酯酶抑制剂能明显改善症状; 血清乙酰胆碱受体抗体和胸腺瘤相关抗体阳性, 低频重复电刺激面神经和腋神经诱发电位波幅呈递减反应, 且纵隔CT检查提示胸腺瘤, 并经手术切除病理证实。另外, 发病3个月后, 又出现多发性神经根神经病的表现: 左侧周围性面瘫, 四肢无力、麻木, 腱反射减弱, 而此时的肌无力经服用胆碱酯酶抑制剂不能明显改善; 肌电图检查发现H反射消失、F反应延长、周围神经运动及感觉传导速度减慢, 提示周围神经及神经根损害; 脑脊液出现明显蛋白-细胞分离现象, 且脑脊液中IgG明显增高, 而血清IgG则正常, 提示椎管内存在免疫过度现象, 而该患者又无脊髓损害的表现, 因而也支持神经根受累。该患者经联合化疗抑制免疫及营养神经治疗后, 病情明显好转, 取得满意疗效。

[收稿日期] 2004-01-08 [修回日期] 2004-03-15
[本文编辑] 曹 静

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