儿童附件扭转的CT特征与临床对照分析

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摘 要：【目的】分析总结儿童附件扭转(AT)的临床特征及CT表现，提高儿童AT的诊断准确率。【方法】回顾性分析经手术病理证实，具有完整临床及CT资料的26例(27灶)儿童AT病例，归纳总结儿童AT的影像特征及病理学基础。根据有无临床症状，分析各种影像征象的阳性率。【结果】26例AT患者年龄为1 d至12岁，平均5.5岁。20例(20/26)以下腹痛为首诊症状。26例中有15例位于右侧(15/26)，10例位于左侧(10/26)，位于双侧者1例(1/26)。25灶(25/27)为完全性扭转，2灶(2/27，7.4%)为不全性扭转。19灶(19/27)合并占位性病变，8灶(8/27)为单纯附件扭转。病灶范围3.4~14.0 cm，平均5.9 cm。12灶(12/27)患侧卵巢体积增大。14灶(14/27)病灶内发现钙化。20灶(20/27)病灶周围发现4~12 mm小囊状结构。增强扫描26灶(26/27)病灶轻度强化(<20 HU)。漩涡征阳性11灶(11/27)，子宫向患侧移位12灶(12/27)，腹水7灶(7/27)，盆腔脂肪间隙模糊11灶(11/27)。CT诊断符合率为88.9%。【结论】胎儿期卵巢囊肿出现囊壁钙化应考虑AT可能。畸胎瘤及卵巢囊肿是儿童AT的危险因素。不对称卵巢体积增大、周围小滤泡是儿童AT特异性较高的CT征象；若同时出现漩涡征、子宫移位、腹水、周围脂肪间隙模糊、畸胎瘤(或囊肿)壁增厚时，应高度怀疑儿童AT的可能。

关键词：附件；扭转；体层摄影术；儿童
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CT Feature and Clinical Analysis of Adnexal Torsion in Children

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Abstract: 【Objective】 To analyze the CT findings and clinical features of adnexal torsion (AT) in children, so as to improve the diagnostic accuracy. 【Methods】 We retrospectively studied the clinical data and CT images of 26 patients (27 lesions) with surgically and pathologically proven pediatric AT. The CT findings and pathological features were summarized. The positive rates of imaging signs were examined. 【Results】 In 26 patients (age range, 1 day-12 years; mean, 5.5 years), 20 complained of lower abdominal pain and 6 were asymptomatic. Fifteen torsions occurred on the right side, ten on the left side and one at both sides. Among the 27 lesions, 25 were complete torsions and 2 partial torsions; 19 were combined with space-occupying lesions and 8 isolated AT. Ranging from 3.4 cm to 14.0 cm, with an average of 5.9 cm, 12 lesions displayed enlarged ovaries on the affected side, 14 calcification and 20 small peripherally cystic structures with 4~12 mm. Contrast-enhanced scan revealed mild enhancement in 26 lesions (<20 HU). 11 lesions presented with whirlpool sign, 12 with uterine deviation to the twisted side, 7 with ascites and 11 with blurred fat space around the pelvic cavity. The diagnostic accuracy of CT was 88.9%. 【Conclusions】 Fetal ovarian cyst with cyst-wall calcification is suggestive of AT. Teratoma and ovarian cysts are risk factors for AT in children. Asymmetrically enlarged ovaries and small peripheral cysts are highly specific CT features of AT. The pediatric AT should be highly suspected in patients with additional imaging

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findings such as whirlpool sign, uterine deviation to the twisted side, ascites, fuzziness of pelvic fat space, and teratoma (cyst) wall thickening.

Key words: adnexal; torsion; computed tomography; children

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儿童及青少年附件扭转(adnexal torsion, AT)发病率为0.02%[1]，占腹痛儿童病例的2.7%[2]，却常常引起卵巢功能/生育能力丧失，乃至威胁生命健康等严重后果[3]。AT发病率低，临床症状不典型，儿童缺乏表述能力，极易导致误诊及漏诊[4]；影像检查成为诊断儿童AT的主要工具和手段，敏感性高于常规体格检查[5]。国内外关于AT影像表现的文献较多，但绝大部分都关注成人AT的影像特征，儿童AT的文献较少[6]。不同年龄阶段儿童AT由于病因不同，影像表现各有特点。本研究回顾性分析经手术病理证实的26例儿童AT的临床及影像资料，与病理结果相对照，分别归纳各种影像征象的病理学基础及阳性率；根据有无临床症状，进一步分析各种影像征象的阳性率，旨在提高儿童AT的诊断水平，最大限度保存患儿卵巢功能及生育能力。

1　材料与方法

1.1　一般资料

收集本院2008年1月至2019年12月经手术病理证实的26例具有完整临床及CT资料的儿童AT病例。本研究通过广州市妇女儿童医疗中心伦理委员会批准。所有患儿家属签署知情同意书。临床资料包括年龄、症状、病程、扭转器官(是否合并肿瘤及肿瘤性质)、手术方式、扭转程度及随访情况。扭转程度分为完全性扭转(扭转大于360°)和不全性扭转(小于360°)。

1.2　检查方法

26名患儿均行下腹部CT平扫及增强扫描。检查不配合者予以0.5 mL/kg体质量10 g/L水合氯醛口服镇静，熟睡后检查。检查采用Philips Brilliance 64排或Toshiba Aquilion 64排螺旋CT机，层厚0.625 mm，层间隔0.625 mm，管电压120 kV，管电流120~200 mAs，矩阵512×512，扫描范围从盆腔入口到耻骨联合下方，若巨大肿块则增大扫描范围直至包括完整肿块。增强扫描采用非离子对比剂(优维显)，剂量为1~3 mL/kg。扫描后原始数据传至后处理工作站，以标准算法重建，重建层厚1 mm，行MPR、VR等后处理。

1.3　图像分析

由2名高年资放射诊断医师双盲法独立阅片。观察及分析内容包括：病灶位置、大小、边界、病灶内成分(钙化/脂肪)、卵巢大小、周围小卵泡及强化程度、漩涡征、子宫位置、腹水及周围脂肪间隙模糊等征象，意见不一致时经双方讨论后达成一致意见。

1.4　数据分析

分析患者的年龄、症状持续时间、病灶大小，患侧卵巢大小，数据记录采用算术均数、$M(P_{25}\sim P_{75})$、最大值及最小值。分别记录病灶发病位置、病灶内是否有钙化/脂肪、周围小卵泡、强化程度、漩涡征、子宫位置、腹水及周围脂肪间隙模糊等征象的阳性率。

2　结　果

2.1　患者基线资料

26例儿童AT算术平均年龄为65.8月，中位年龄72月(11.8~96.0月)，范围1 d至12岁。20例以下腹痛为首诊症状，6例无临床症状。26例(27灶)行开腹或腹腔镜手术，其中15例的病灶位于右侧，10例的病灶位于左侧，双侧病灶者1例(1/26)。19灶合并占位性病变，8灶为单纯附件扭转。25灶完全性扭转行患侧病灶切除术，2灶不全性扭转行卵巢肿物剔除术。术后随访3个月到10年，平均随访时间5.1年，至本文撰写为止，未见复发病例(表1)。

2.2　患者CT表现

27灶(26例)病灶范围为3.4~14.0 cm，算术平均直径5.9 cm，中位直径5.7(4.1~6.9) cm。12灶患侧卵巢体积增大(12/27)，14灶病灶内钙化灶(14/27)，20灶周围发现4~12 mm小囊状结构(小卵泡)

(20/27)。增强扫描26灶病灶轻度强化(<20 HU)(26/27)。漩涡征阳性11灶(11/27),子宫向患侧移位12灶(12/27),腹水7灶(7/27),周围脂肪间隙模糊11灶(11/27)。1灶AT梗死后病灶钙化,2灶胎儿期囊肿AT术前未能准确诊断,CT诊断符合率为24/27。

2.3 各组患者临床和影像学表现

根据有无临床症状,笔者把儿童AT分为无症状组AT及症状组AT。在症状组AT,进一步根据AT是否合并占位性病变分为:①占位合并AT组;②单纯AT组。

2.3.1 无症状组 全部为胎儿期卵巢囊肿扭转,共6例(6灶)。患儿年龄1 d至6个月,算术平均年龄为1.8月,中位年龄1.2(0.1~2)月。6(6/6)例无临床症状。病灶位于左侧4例,右侧2例。4例(4/6)完全性扭转及1例(1/6)卵巢囊肿自截行患侧病损切除术,1例(1/6)不全性扭转行囊肿剔除术。CT表现:6例(6/6)均表现为腹部单房囊肿,1例(1/6)囊壁周围见少量正常卵巢组织。囊肿范围2.3~6.0 cm,算术平均值4.0 cm,中位值3.6(3.1~5.1)cm。囊液密度均匀5例(5/6),1例(1/6)出现液-液平面,囊液CT值14~78HU。囊壁厚薄均匀,5例出现点、线状钙化(图1),增强扫描囊壁轻度均匀强化。

2.3.2 症状组 患儿年龄3个月至12岁,算术平均年龄85月,中位年龄96(69~99)月,共20例(21灶),所有儿童均出现临床症状。

占位合并AT:共13例(13灶)。患儿年龄3月至12岁,算术平均年龄84.6月,中位年龄84(60~120)月。13例(13/13)以下腹痛为首诊症状,病程1~14 d,症状算术平均持续时间4.8 d,中位时间4(3~7) d。其中5例(5/13)伴恶心、呕吐,1例(1/13)伴发热,1例(1/13)伴便秘。病灶位于右侧10例,左侧3例。合并畸胎瘤10例(10/13)且全部为初潮前女童,合并卵巢囊肿3例(3/13)且全部为初潮后女童。1例畸胎瘤合并副中肾管囊肿及1例对侧输卵管副中肾管囊肿,副中肾管囊肿一并切除。此2例副中肾管囊肿直径均<0.5 cm,CT图像上无法辨认。12例(12/13)为完全性扭转行病灶切除,1例(1/13)为不全性扭转行畸胎瘤剔除术。CT表现:病灶3.7~14.0 cm,平均直径7.1 cm,中位直径6.7(5.7~7.6) cm。12例(12/13)病变直径>5 cm。8例(8/10)畸胎瘤与患侧卵巢分界不清,瘤壁增厚(图2),病灶内可见脂肪及钙化,其中1例(1/10)显示脂-液平面。5例(5/13)表现为单房囊性肿物(图3),囊壁与患侧卵巢部分分界清晰;其中2例(2/5)为囊性畸胎瘤,3例(3/5)为生理性囊肿。13例病灶周围探及直径4~12 mm小囊样结构(卵泡),增强扫描囊壁轻度强化。漩涡征阳性8灶,子宫向患侧移位6灶,腹水4灶,盆腔脂肪间隙模糊8灶。

单纯AT:共7例(8灶)。患儿年龄3~9岁,算术平均年龄为85.7月,中位年龄96(84~96)月。7例(7/7)以下腹痛为唯一症状,病程3~10 d,症状算术平均持续时间5 d,中位时间4.0(3.5~5.5) d。病灶位于双侧者1例(1/7),病灶位于左右侧者各3例(3/7)。1例(1/7)为单纯卵巢完全性扭转(合并对侧卵巢梗死后钙化)(图4),6例(6/7)为卵巢和输卵管完全性扭转(图5)行病灶切除术,无单纯输卵管扭转病例。CT表现如下:病灶3.4~14.0cm,平均直径5.4 cm,中位直径5.6 (4.3~6.4)cm。1灶患侧卵巢陈旧梗死并钙化。7灶显示患侧卵巢体积增大,直径2.5~6.7 cm,平均直径5.1 cm,中位直径4.7(3.8~6.1)cm。7例病灶周围探及直径4~12 mm小囊样结构(卵泡),增强扫描囊壁轻度强化。漩涡征阳性3灶,子宫患侧移位6灶,腹水3灶,盆腔脂肪间隙模糊3灶。

3 讨 论

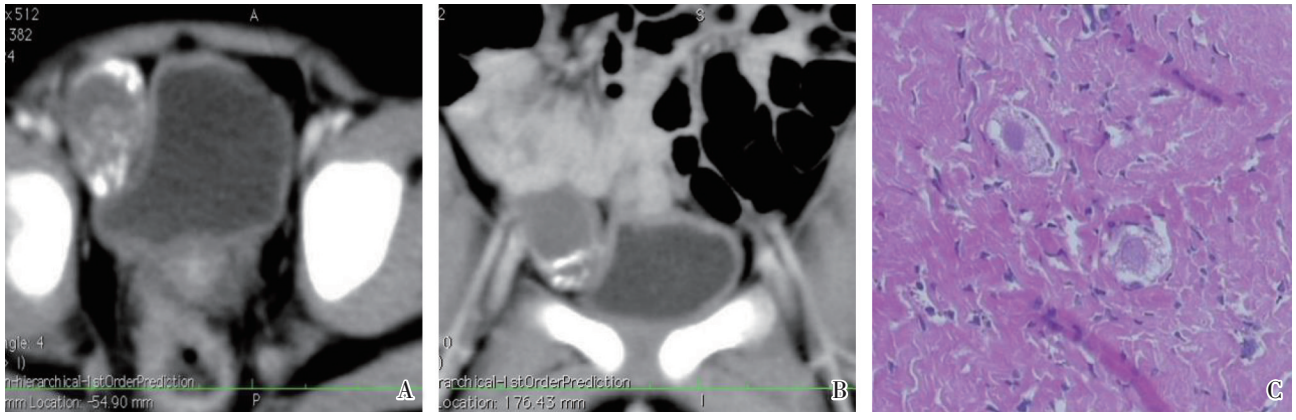
3.1 概 述

AT指包括卵巢、输卵管和/或其占位性病变的扭转。子宫及卵巢体积小、周围韧带松弛、输卵管长[7]、突发剧烈活动导致腹内压增高[8]都是儿童AT的重要诱因,占全部AT的15%[9]。儿童AT平均发病年龄为9.2岁[10],表现为突发性右下腹痛[11]且疼痛持续时间小于48 h[12],伴呕吐、发热。本研究儿童AT具有以下特点:①发病年龄早:除胎儿期囊肿扭转病例外,本研究AT平均发病年龄为7.1岁。这可能与本院为专科医院,病人选择性偏倚导致。②病程长:平均病程4.9 d,最长14 d。疼痛持续时间大于10 h与扭转组织坏死率增加有关[13],这可能是大部分病例(25/27)无法保留患侧卵巢的原因之一。③辅助检查不及时:影像检查可提高诊断准确性,早期保守治疗可以保留卵巢功能及未来的生育能力[14]。笔者建议下腹痛而无其他消化道症状,疼痛持续时间大于48 h的女童应尽早行影像检查;若发现单侧附件肿大,应高度怀疑AT的可能[15]。

表1　26例27灶儿童AT临床及CT影像表现

Table 1　Clinical and CT imaging manifestations of AT in 26 children with 27 lesions　[n/N, M (P_{25}~P_{75})]

Groups	Asymptomatic AT	Symptomatic AT	
		Combining with space-occupying lesion	Isolated AT
n	6/27	13/27	8/27
Clinical features			
Median age/months	1.2(0.1~2.0)	84(60~120)	96(84~96)
Symptom	NA	Abdominal pain, vomiting, fever, constipation	Abdominal pain
Duration/days	NA	4.0(3.0~7.0)	4.0(3.5~5.5)
Location/R	2/6	10/13	4/8
CT manifestations			
Asymmetric enlarge ovarian	NA	5/13	7/8
Peripherally cystic structures	NA	13/13	7/8
Calcification	5/6	8/13	1/8
Mildly enhanced	6/6	13/13	7/8
Whirlpool sign	NA	8/13	3/8
Uterus deviation	NA	6/13	6/8
Ascites	NA	4/13	3/8
Bur fat around	NA	8/13	3/8



Female, 6 months old, asymptomatic. The ovarian cyst was twisted and infarcted in the prenatal period. A and B: A round-shaped cystic lesion was on the right side of the bladder, with multiple punctate and linear calcifications on the cyst wall and uniform thickness. The enhanced scan showed the wall of the cyst was with slight line-like enhancement. C: Microscopically, the right ovarian tissue was diffused with coagulative necrosis and scattered with flaky calcium salt deposits (HE staining ×100). Pathological diagnosis: hemorrhagic necrotic infarction of the right ovary.

图1　无症状AT患儿影像学和病理学特征

Fig. 1　Imaging and pathological features of children with asymptomatic AT

④复发率低：5%~18%儿童AT出现复发[4]。本研究暂未见复发病例，推测与病灶全部切除有关。

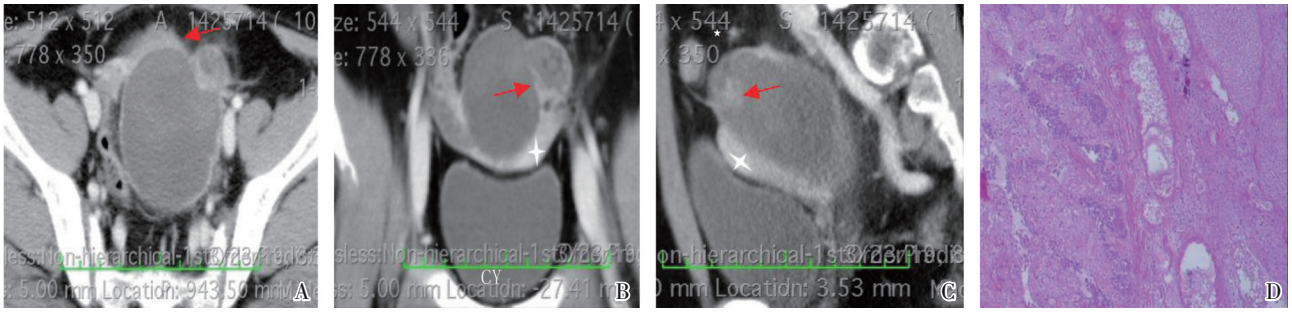
3.2　儿童AT的临床表现与影像特征

外科手术是AT诊断的金标准，手术可以明确

Female, 10 years old, with abdominal pain and fever for 3 days. Left ovarian teratoma was torsion and infarction. A: A soft tissue mass was located on the left-back of the bladder, with a whirlpool sign(thick red arrow). Noted that the contralateral ovary was normal(white star). B: Multiple small peripherally cyst was found around the lesion(thin red arrow). C: Hemorrhage and necrosis of the cyst wall were showed by the microscope, part of the cyst wall was covered with squamous epithelium single cubic epithelium. Hair follicles, sweat glands, keratinocytes, and sebaceous gland tissues were seen in the lesion. Pathological diagnosis: left ovarian teratoma and the ovarian tissue was with hemorrhagic infarction(HE staining ×40)

图2 占位(畸胎瘤)合并AT患儿影像学和病理学特征

Fig. 2 Imaging and pathological features of children with AT combining with space-occupying lesions (Teratoma)

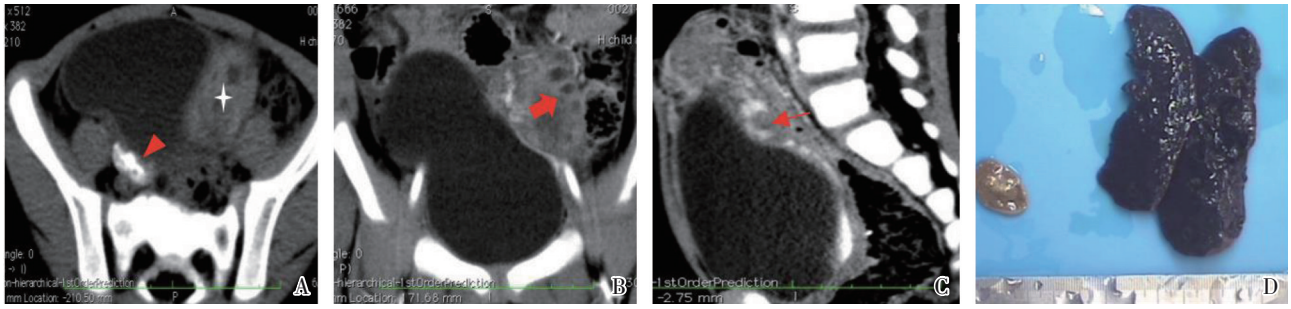


Female, 9 years old, with abdominal pain for 10 days. Left ovary cyst (CY) combining with the left ovary and the fallopian tube was twisted and infarcted. A: The peripheral fat around the CY was blurred (red arrow). B: The uterus (white star) was shifted to the left. The wall of the cyst between the left ovary and the CY (red arrow) became thicker. Noted that the right ovary was normal. C: The wall of the cyst (white arrow) was thicker. D: The CY and ovary were dark brown during the operation. Diffuse hemorrhage and necrosis of the ovary and fallopian tube were found by the microscope, with a small amount of inflammatory cell infiltration (HE staining ×40). Pathological diagnosis: diffuse hemorrhage and necrosis of the left ovary.

图3 占位(卵巢囊肿)合并AT患儿影像学和病理学特征

Fig. 3 Imaging and pathological features of children with AT combining with space-occupying lesions (Ovarian cyst)

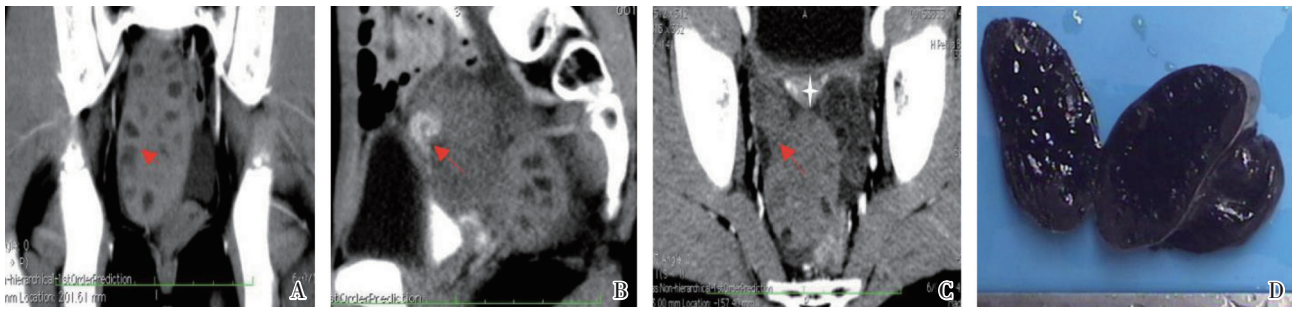
是否扭转并评估附件活性,但需要根据临床诊断来决定是否需要手术[16]。超声是临床疑诊儿童AT首选的影像学检查方法,诊断准确率为74%[17],假阴性发生率45%~61%[18],超声检查容易受操作者水平的影响。MRI软组织分辨率高,无电离辐射,诊断符合率为85.7%[19],但较难应用于急诊。CT作为儿童腹痛的一种常见的诊断手段,诊断准确率与超声相当[20],本研究为24/27。卵巢体积增大是AT的常见征象,阳性率为44.4%(12/27),由静脉扭转导致的卵巢间质充血水肿所致。患侧卵巢体积大于对侧3倍以上[21]或>1岁儿童卵巢直径>5 cm[10]即为阳性。卵巢间质水肿使液体外渗进入卵泡,表现为卵巢周边直径8~12 mm小囊影,发生率约74%[22],也是AT常见征象之一。约21例(21/27)的患侧卵巢平扫密度增高(平均CT值56 HU),笔者推测可能与卵巢组织少量渗血有关。钙化的阳性率为51.9%(14/27),不同年龄阶段AT钙化的病因及形态不同。卵巢由卵巢动脉及子宫动脉双重供应,故即使病灶出现强化亦不能排除梗死的发生[23],本研究中的26病例(26/27)出现患侧卵巢轻度强化。90%的AT患者病灶旁可见直径>10 mm的不规则/管状、绳索状肿块,向子宫方向延伸,此为"漩涡征",由扭曲的血管蒂或输卵管组成,是AT高度特异性征象,但阳性率仅40.7%[24]。



Female, 10 years old, with abdominal pain for 3 days. Bilateral isolated ovarian torsion and infarction. A: Calcification lesion was found on the right posterior side of the bladder (red triangle), which was confirmed to be calcification after right ovarian infarction. The left side of the bladder showed a slightly high-density soft tissue lesion, with multiple small peripherally cyst (white star). B: Multiple small vesicles around the lesion (thick red arrow). C: " Whirlpool sign " was showed (thin red arrow). D: Gross specimen showed calcium deposits in the right ovary. The left ovary was enlarged with a dark brown surface. Pathological diagnosis: diffuse hemorrhage and necrosis of bilateral ovarian tissue, local fibrous tissue hyperplasia of the right ovary with focal calcium deposition.

图4　单纯AT患儿影像学和病理学特征

Fig. 4　Imaging and pathological features of children with isolated AT



Female, 8 years old, with abdominal pain for 3 days. Isolated right ovarian infarction. A: The volume and the CT density of the right ovary increased, with multiple small peripherally cyst (red arrow). B and C: The twist vascular pedicle of the right side showed a “whirlpool sign” (red arrow), and the uterus (white star) was slightly shifted to the right. D: The right ovary was enlarged and dark brown. Pathological diagnosis: hemorrhagic infarction of the right ovary.

图5　单纯AT患儿影像学和病理学特征

Fig. 5　Imaging and pathological features of children with isolated AT

AT发生后，卵巢悬韧带及固有韧带牵拉子宫向扭转侧骨盆移位，周围炎性渗出等导致子宫向患侧移位，并有腹水及周围脂肪间模糊等非特异性征象。

笔者发现无临床症状的儿童AT，由于诱因不同，影像表现与症状组不同，具有自己的特点，不能一概而论。根据有无临床症状，我们把儿童AT分为2组：①无症状组；②症状组。在症状组，占位性病变作为儿童AT的重要影像特征，可影响AT其他影像征象的阳性率。为此，我们进一步把症状组AT划分为：占位合并AT组及单纯AT组，现分别叙述如下：

3.2.1　*无症状组*　16%的儿童AT发生在1岁以下[10]。胎儿期卵巢从腹部下降使卵巢活动度增加[25]，母体激素刺激导致胎儿卵巢增大和囊肿形成等因素是胎儿期AT的重要诱因。本组AT多无临床症状，多为影像检查偶然发现。AT自截罕见，被描述为腹部自由漂浮的钙化或非钙化肿块[26]，本组1例。出血坏死性囊肿是本组AT独特的病理学改变，包括囊壁钙化、肉芽肿反应、含铁血黄素沉积和纤维化[27]，这些改变提示扭转可能发生在宫内。此组AT的影像特征为囊肿壁点、线状钙化；囊液密度均匀，增强扫描囊壁均匀强化不明显[28]。由于扭转可能发生在宫内，其他征象多为阴性。

3.2.2　*症状组*　以下腹痛为主要临床表现，诊断主要依靠影像检查，诊断及鉴别诊断困难。

占位合并AT：受累卵巢体积增大、重量增加，

使其容易成为扭转的支点,特别是当占位的直径>5 cm[10]时。畸胎瘤和出血性/滤泡性囊肿占儿童AT的51%~84%[24],是儿童AT最常见的诱因[29];恶性肿瘤侵犯/粘连周围结构,反而不容易出现扭转,仅占AT的2%[30]。不同年龄阶段合并占位的性质也不同:畸胎瘤是初潮前AT的重要诱因,生理性囊肿则为初潮后。形态规则、边缘清晰的含钙化、脂肪、脂-液平面的肿块是畸胎瘤的影像特征。当畸胎瘤边缘不清,"壁"增厚并强化时,应警惕儿童AT的发生。初潮后第一个月约20%的卵巢会出现囊肿[7],大部分AT发生在此时,且93.3%[31]的病例会出现囊壁增厚。扭转早期,囊壁增厚可不明显;随着扭转程度加重,扭转蒂附近的囊壁可出现局限性增厚乃至弥漫性增厚,且厚薄不均,最厚处一般位于扭转蒂附近,增强后囊液及囊壁一般无强化。输卵管旁囊肿(副中肾管囊肿/卵巢旁囊肿)是儿童输卵管扭转最常见的原因[32]。本研究无单纯输卵管扭转病例,2例副中肾管囊肿均为术中偶然发现,且直径均<5 mm。除不对称卵巢体积增大的阳性率低,仅为5例(5/13)外,其他影像征象的阳性率高且典型。

单纯AT:25%~68%的儿童AT发生在正常的附件[7, 14]。本组AT临床症状单一(腹痛),病程长(平均5 d),鉴别诊断难度最大。但本组AT的影像表现典型,除漩涡征外,其他征象的阳性率也最高。张成周等[33]认为,漩涡征与扭转的圈数、累及长度及发病时间长短、扫描层面有关。笔者认为漩涡征的发现与扫描层厚有关,层厚越薄,通过多平面重建等后处理技术,越容易发现扭转的血管蒂。早期影像检查是本组患儿确诊的关键,对挽救本组患儿的生育能力至关重要。

3.3 鉴别诊断

无症状组AT需要与胎粪性腹膜炎、肠重复畸形相鉴别。卵巢囊肿是女胎儿最常见的腹部囊性包块。胎粪性腹膜炎与AT在影像上均表现为囊性包块及囊壁点、线状钙化。但前者多合并消化道异常,病灶周围脂肪间隙多模糊不清;后者多无症状,病灶周围脂肪间隙清晰。肠重复畸形很少出现钙化,且病灶多与相邻肠管平行,超声检查可探及囊肿内高回声黏膜结构,AT无此特征。症状组AT鉴别诊断困难。笔者认为,对持续性下腹痛女童应尽早行影像学检查,若出现一种或几种典型AT征象时,尤其是合并巨大占位时应尽早行腹腔镜/手术探查,尽量保留女童卵巢功能。

AT是儿童腹痛的少见病因,容易出现误诊及漏诊。具有胎儿期腹部囊肿的女婴若发现囊壁点、线状钙化应考虑AT的可能。初潮前合并畸胎瘤、初潮后合并囊肿是AT的危险因素,尤其是当肿块直径>5 cm时。不对称卵巢增大并周围多发小滤泡是儿童AT的影像特征,同时合并漩涡征、子宫患侧移位、腹水、周围脂肪间隙模糊、畸胎瘤(囊肿)壁增厚时,应高度怀疑儿童AT的可能。

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