

· 综述 ·

继发于 Perthes 病的成人扁平髋：1 例报告和综述[△]朱高明¹, 郭艳波^{1,2}, 张加豪¹, 李刚^{2*}

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摘要: Perthes 病 (Legg-Calvé-Perthes disease, LCPD) 是一种特发于儿童的以股骨头坏死为特征的罕见性疾病, 病因不明, 起病隐匿。随着病程进展, 易发展为扁平髋甚至合并严重的髋关节骨性关节炎, 头臼匹配差甚至髋臼反倾, 常常不得不接受全髋关节置换术, 治疗难度大。本文报道 1 例继发于 LCPD 的成人扁平髋骨性关节炎患者, 并对围绕 LCPD 相关的研究进展进行综述。

关键词: Perthes 病, 扁平髋, 髋臼反倾, 人工髋关节置换

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Adult coxa plana secondary to Legg-Calvé-Perthes disease: a case report and literature review // ZHU Gao-ming¹, GUO Yan-bo^{1,2}, ZHANG Jia-hao¹, LI Gang². 1. The First Clinical College, Shandong University of Traditional Chinese Medicine, Jinan 250014, China; 2. Affiliated Hospital, Shandong University of Traditional Chinese Medicine, Jinan 250011, China

Abstract: Legg-Calvé-Perthes disease (LCPD) is a rare disease characterized by avascular necrosis of the femoral head that occurs specifically in children, with unknown etiology and insidious onset. With the progression of the disease, it is prone to develop into coxa plana or even severe hip osteoarthritis in adulthood due to poor head and acetabulum matching or even acetabular inversion. The adult with coxa plana secondary to LCPD is difficult to treat, and often have to accept total hip arthroplasty. This article reports a case of adult coxa plana and severe osteoarthritis secondary to LCPD, and reviews the research progress related to LCPD.

Key words: Legg-Calvé-Perthes disease, coxa plana, acetabular inversion, total hip arthroplasty

Perthes 病 (Legg-Calvé-Perthes disease, LCPD) 是一种好发于儿童的以股骨头坏死为主要特征的髋关节自限性疾病^[1]。该病临床相对少见, 只有 0.5~30/10 万的发病率, 多为单侧发病, 双侧发病率仅为 9.4%, 总体病程约为 3~4 年^[2, 3], 其中, 多发于 4~8 岁儿童, 最新流行病学调查显示, 发病年龄在 8 岁以下儿童占比高达 70.2%^[4]。虽然该病为自限性疾病, 由于该病正处于骨骼快速发育期, 在后期恢复过程中, 患侧股骨头的异常塑形, 干骺端病变, 导致股骨近端发育不良, 股骨头形态不规则, 严重影响髋关节功能, 极大的增加了成年后扁平髋、关节退行性变等风险^[5]。本文报道 1 例继发于 LCPD 的成人扁平髋骨性关节炎患者, 并根据文献对该病相关的研究进展进行综述。

1 病例报告

患者, 女, 33 岁。自小行走后即被发现右下肢跛行, 渐重, 至当地医院就诊, 诊为“右侧发育性髋关节脱位”, 给予铝板外展位固定, 9 个月后拍片复查, 见股骨头坏死, 拆除固定支架后未予特殊处理。患者遗留右下肢跛行, 右髋行走后偶感疼痛, 于 2003 年 9 月明显加重, 遂来本院就诊, 经查体、拍片, 诊为“右侧扁平髋”, 收住入院拟行截骨术或人工关节置换术。经院内骨科会诊讨论, 一致认为人工全髋关节置换术为较合适的治疗方法, 其余方法均不能全面解决患者疼痛、跛行、下肢短缩等问题, 但鉴于患者年龄较小, 暂不宜行关节置换, 建议外院会诊后决定治疗方案, 停手术后出院。出院后患者未行进一步治疗, 2022 年 11 月再次于本院就诊, 诉“右髋疼痛伴活动受限加重 3 个月余”, 专科检查: 右下肢跛行, 右髋行走后疼痛, 站立位时患肢呈外旋及轻度内收位, 骨盆向患侧倾斜, 右侧臀肌萎缩明显, 臀沟

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比左侧下降, Trendelenburg 征 (+)。仰卧位, 股骨大粗隆顶点高于 Nelaton 线, 右髋关节外展、内旋、屈曲及后伸等各向活动均明显受限。影像检查所见见图 1a~1c。结合病史、查体及影像学检查, 诊断为“右侧扁平髋 (Perthes 病)”。完善检查后, 于腰麻下行右侧人工全髋关节置换术。具体过程如下: 麻醉成功后, 患者侧卧位, 术区消毒、铺巾, 取右髋后外侧入路, 切口长约 15 cm, 依次切开, 顺肌纤维方向钝性分离臀大肌, 于股骨大粗隆止点处切断部分外旋肌群肌肉, 切开发节囊, 使股骨头脱位, 见股骨头明显变扁, 股骨颈短缩, 关节软骨剥脱, 软骨下骨裸露, 保留骨矩约 1.5 cm 截骨; 进一步显露髋臼, 去除臼内增生结缔组织, 见关节软骨磨损严重, 髋臼周围硬化增生, 保持外倾 45°、前倾 15°用髋臼锉逐级

扩大髋臼, 至露出软骨下骨新鲜渗血止, 保持上述角度压配式置入直径 48 mm 生物型髋臼金属杯, 2 枚 20 mm 髋臼螺钉辅助固定, 安装陶瓷内衬, 臼杯前外侧裸露区域予以颗粒植骨; 屈曲内收患肢, 逐级扩髓, 保持前倾 15°打入 8 号生物型假体柄, 安装 32 mm S 陶瓷球头, 复位后测试髋关节稳定性及灵活性可。冲洗伤口, 仔细止血, 放置引流, 缝合关节囊及外旋肌群, 逐层关闭切口, 敷料包扎。术后患者述患侧足底及足根部麻木不适, 下肢肌力及活动可。术后第 3 d 开始持双拐下地活动。随访 4.5 个月, 患者恢复良好, 疼痛和麻木不适逐渐缓解, Harris 功能评分 93.5。影像复查见右髋旋转中心恢复良好, 假体位置良好 (图 1d)。

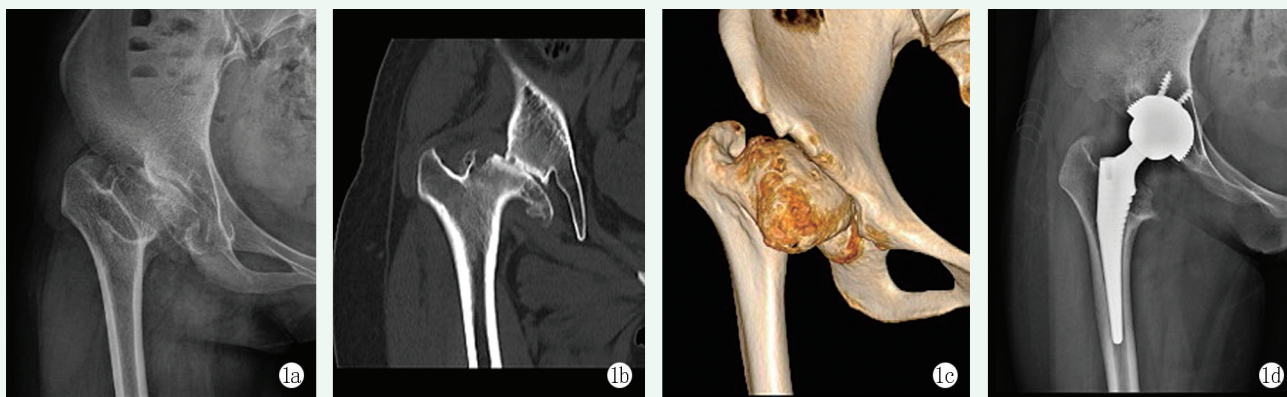


图 1. 患者女性, 33 岁, 因右侧扁平髋 (Perthes 病) 行右侧人工全髋关节置换术。1a: X 线片示右侧髋臼浅, 股骨头变扁, 股骨颈遮挡显示欠清, 右侧沈通氏线欠连续; 1b, 1c: CT 及三维重建示右侧髋臼窝浅平, 右侧股骨头变扁、不规则并向外上方移位, 右侧髋关节间隙变窄; 1d: 术后 2 个月 X 线片示假体位置良好。

Figure 1. A 33-year-old female who has undergone a right total hip arthroplasty due to Perthes disease affecting right hip. 1a: Preoperative X-ray showed a shallow acetabulum on the right side, with flattening of femoral head, lack of clear occlusion of femoral neck, and lack of continuity of Shenton's line on the right side; 1b, 1c: Preoperative CT and 3D reconstruction showed a shallow and flat acetabular fossa on the right side with flattened, and irregular femoral head shifting outward and upward, and the narrowed right hip space; 1d: Plain radiographs 2 months after surgery showed that the prosthesis was in good position.

2 讨论与文献综述

目前关于 LCPD 病发病原因尚不明确, 现有假说认为遗传、机械应力改变、全身性条件如肥胖、多动症等是导致该病的重要因素, 其中, 较为主流的观点认为重复的机械应力干扰了骨髓的正常血供^[6]。除此之外, 高凝状态也是一个重要的致病因素, 有研究显示凝血因子 V 突变、G20210A 凝血酶原突变、因子 VIII 水平升高和蛋白 S 缺乏可能是导致 LCPD 发病的重要因素, 但该论点一直存在争议, 目前为止没有得到确切的证实^[7]。

股骨头骨髓坏死大致经历 4 个病理过程: 缺血

坏死、碎裂、再生、愈合。在坏死阶段, 坏死骨中钙的增加使股骨头更容易受到微小损伤, 机械应力改变, 关节间隙增加, 继发软骨下骨折。随后, 骨细胞死亡增加, 微骨折形成, 在血运重建阶段, 坏死骨被吸收, 进一步损害股骨头机械性能^[8]。一直以来, 对于 LCPD 的诊断、分期、治疗方式的选择及预后评估都是以 X 线片作为首选, 早期 X 线片可显示典型的骨质硬化、囊性改变和“新月征”等表现; 塌陷后, 晚期可出现股骨头球形丧失和退行性关节炎等影像学表现^[9]。基于此衍生了 Catterall 分型、Herring 分型、Salter-Thompson 分型等反应股骨头受损程度的分型方式, 帮助疾病的诊断及治疗。但对早中期该类型的患者, 普通的 X 线片无法精确

展示动态的血运情况, MRI 或增强 MRI 在股骨头灌注指数、突出指数、受累程度及分期等具有更重要的评估意义, 典型表现为骨髓水肿、股骨头骨骺的信号变化以及双线征等直接征象^[10, 11]。

对于儿童 LCPD 患者的治疗, 目前还没有确切的可以改善骨改变的治疗方法, 减轻负重及手术方式仍是当前最主要的策略, 得到了广泛应用。保守治疗, 如改善髋关节活动范围, 仍然是基本的治疗标准, 通常以预防股骨头畸形, 改善患儿临床症状为目标。对于年龄<8 岁、病灶位于股骨头前部且范围<50%, 或仅表现为骨密度轻度改变及 Catterall 分型在 I、II 级或 Herring A 型的早中期患儿, 目前其治疗理念主要以保守治疗为主, 包括制动休息、石膏或支具外固定等方式, 根据症状使用止痛、抗炎类药物^[12, 13]。对于病情严重、股骨头骨骺病变范围>50%的患儿, 单纯保守治疗已不足以缓解其临床症状及阻止后期的严重并发症, 常采用手术治疗。其中, 年龄>6 岁, Catterall 分型为 I、II 型或 Herring 分型在 B-C 型的患儿, 可采用 Salter 骨盆截骨术或联合股骨截骨术、骨盆三联截骨术等包容性手术方式。针对典型的 LCPD 后畸形, 头臼匹配尚可, 股骨颈缩短、粗隆上移、无晚期骨关节炎的年轻患者, 通常采用外科脱位技术使股骨颈延长, 改善机械应力, 在此基础上, 股骨头扁平、头臼匹配度较差者, 可选用股骨头缩小成形术来维持股骨头球形形态, 改善疼痛症状及头臼匹配度, 延缓疾病进展^[14-16]。除此之外, 根据疾病进展状态, 可适当选用股骨头表面重建术等保髋手术, 防止或延迟成年后的髋关节置换。

就预后而言, 发病时的年龄和严重程度是决定 LCPD 患者预后的主要因素^[17]。一般情况下, 发病年龄≤6 岁的患儿, 预后更好^[18]。目前临床主要采用 Stulberg 分型评价 LCPD 患者的预后结局^[19], 主要分为 I 型: 股骨头及髋臼形态正常, 头臼正常匹配; II 型: 股骨头为球形, 且具有股骨近端骨骺增大、股骨颈短、髋臼异常陡峭 1 个或以上的特征; III 型: 股骨头非球形但不扁平的卵球形, 颈臼改变, 头臼椭圆匹配, 具有 II 型所述的 3 种异常中的至少 1 种; IV 型: 股骨头是平的, 但由于髋臼重塑, 股骨头是全等的, 有 II 型所描述的畸形中的 1 种或以上; V 型: 股骨头扁平或平坦, 髋臼无适应性改变, 头臼线性不匹配, 由于没有髋臼重塑, 与正常股骨颈不一致^[18, 20]。I、II 型髋关节头臼吻合, 一般不会发生骨关节炎, 整体预后良好。Froberg 等^[21]对 167 例非手术治疗的 LCPD 患者平均随访时间为 47 年的研究及

发现, I/II 型股骨头患者的髋骨关节炎患病率为 2%。最新文献报道, 对于 7 岁以下的儿童行减轻负重治疗, 88.3%的患儿取得了良好结果^[22]。III 型和 IV 型髋关节球面匹配度降低, 可引起轻中度骨关节炎^[19]。若失治误治, 随着病程进展, 股骨头骨骺愈合扁平变宽, 髋臼适应性变浅增大, 呈现扁平髋畸形, Stulberg 分型属 IV 型或 V 型, 此类型患者预后差, 致残率高。头臼扁平畸形致髋关节疼痛, 下肢短缩、臀腿部肌肉萎缩致跛行、活动受限, 骨盆脊柱出现继发性改变, V 型的患儿髋关节可在 50 岁前发展为重度骨关节炎, 针对此阶段患者, 临床可见零散报道, 通过股骨头表面重建术、联合截骨术、股骨头缩小成形术等保髋手术干预, 以改善股骨头球形和髋臼匹配性及稳定性, 避免肢体短缩和髋臼发育不良, 改善生活质量, 尽可能推迟进行人工全髋置换手术的时间, 临床随访发现, 其短、中期影像学 and 临床效果总体满意^[23-26]。

伴随着成年, 部分患儿髋部退变为中至重度骨关节炎, 此时患者遗留有严重的后遗症。成年 LCPD 病后遗症患者与年龄和性别匹配的对照组相比, 功能、身心健康状况更差, 常常需要在年轻时就不得已行全髋关节置换术^[21, 27]。研究显示, 成年之后约有 29.3%受访者经历至少一次手术, 而最常见的手术选择即全髋关节置换术^[4]。除部分患者髓腔狭细或前倾角过大外, 使用常规的生物型髋关节假体^[28], 即股骨侧通过单体式近端固定柄重建即可获得满意的临床疗效。全髋关节置换围手术期并发症多发, 远高于普通患者^[29], 如假体周围骨折、坐骨神经损伤、下肢不等长等直接影响康复进程、满意度和功能结局。既往髋部有手术史与并发症发生存在相关性^[28]。坐骨神经损伤大部分是源于全髋关节置换术后肢体长度恢复牵拉所致, 研究统计, 发生坐骨神经损伤病例平均肢体延长 1.9 cm^[28]。事实上, 大量的中长期随访研究已证实全髋关节置换术是治疗 LCPD 病所致的扁平髋的有效措施, 10 年生存率高达 90%^[30-34]。

需要特别注意的是, 髋臼反倾在 LCPD 病中发生率高达 31%~60%^[35], 甚至远远高于髋关节发育不良患者。一项针对 40 岁以下接受全髋关节置换患者的研究发现, 髋臼反倾发病率高达 1/3^[36]。髋臼反倾容易造成髋臼撞击, 过早引发髋关节炎^[36], 与 LCPD 病预后不良相关。单侧发病的患者双侧髋臼可能都会表现出髋臼反倾, 并且预后更差。在影像学评估中, 通常通过髋关节正位 X 线片上的阳性“交叉征”、“坐骨棘征”等来评估, 但一致性和重复性并不

好^[37]。有学者通过核磁的轴位像测量髋臼前倾角进一步证明了“交叉征”与影像学的严重程度、治疗的预后无关^[38]。国内有学者尝试通过断层CT扫描为髋臼反倾的评估建立新的评价参数,并推测其是半骨盆远端后倾所致^[39]。在全髋关节置换术干预时,髋臼反倾则会导致臼杯后壁缺乏骨性覆盖,假体与宿主接触面积减少,影响假体的稳定性,需要引起临床关注^[36]。对于此类技术要求高的复杂全髋关节置换术,要充分做好术前规划,评估股骨近端及髋部的解剖结构,合理选择组配式或定制假体,运用计算机导航或机器人辅助技术^[40],在保证稳定的前提下,尽可能恢复双下肢等长,以提高临床疗效和满意度。

本例患者幼年发病,当地医院误诊为“右侧发育性髋关节脱位”予以支架外展位固定,9个月后复查X线片提示有股骨头坏死。患者家属未予重视,未得到及时诊治以改善头臼匹配,后患肢逐渐出现疼痛、跛行、下肢短缩症状;进入青春期后又于本院就疗,综合考虑患者病情及手术风险,患者未行手术干预,随着成年和病情进展发展为扁平髋并髋骨关节炎。治疗上予常规生物型假体全髋关节(沃尔德马林克),股骨侧髓腔狭细,在保证力学稳定前提下选择小号生物柄以防医源性骨折,髋臼侧并无髋臼反倾,前方缺损明显,无骨性覆盖,臼杯边缘外露明显,以髋臼螺钉增强髋臼侧稳定性,以自体松质颗粒骨植入、压实增加前壁完整性;术后肢体长度恢复,虽并发坐骨神经牵拉损伤,但症状随着康复进程逐步消失,长期随访中,患者未诉不适,临床疗效满意。

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