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## • 病例报告 •

## 慢性骨髓炎继发进展型巨大股骨远端表皮样囊肿 1 例

季卫锋, 童培建, 马镇川, 倪桂宝, 沈国华, 周海龙, 姚晓冬, 肖鲁伟  
(浙江中医药大学附属第一医院骨伤科, 浙江 杭州 310006)

关键词 骨髓炎; 股骨; 表皮囊肿

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**Progressive huge epidermoid cyst of distal femur in chronic osteomyelitis: a case report and review of literature** Ji Wei-feng, TONG Pei-jian, MA Zhen-chuan, NI Gui-bao, SHEN Gou-hua, ZHOU Hai-long, YAO Xiao-dong, XIAO lu-wei. Department of Orthopaedics, the First Hospital Affiliated to Zhejiang University of Traditional Chinese Medicine, Hangzhou 310006, Zhejiang, China

**KEYWORDS** Osteomyelitis; Femur; Epidermal cyst

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患者, 男, 60 岁, 因“左下肢肿胀疼痛 30 余年, 加重 1 年余”入院。42 年前因在水库洗澡后回家发现左下肢肿胀疼痛, 去当地医院就诊, 诊断为左股骨骨髓炎行手术治疗, 慢性骨髓

炎反复发作, 经抗生素治疗后缓解。近年来病情加重, 左大腿外侧出现窦道, 流脓。2009 年 1 月在当地医院摄 X 线片示: 左股骨远端硬化和透亮线, 边界不清, 并见骨膜反应(图 1a)。CT 示软组织扩张, 皮质破坏(图 1b)。行左股骨干骨髓炎钻孔减压术。2009 年 5 月肿痛复发在当地医院就诊, MR 示股骨远端

通讯作者: 季卫锋 E-mail: jiwefeng@zjtcn.net



**图 1** 男,60 岁,慢性骨髓炎 **1a.** 2009 年 1 月 X 线片示左股骨远端硬化和透亮线,边界不清,并见骨膜反应 **1b.** CT 示软组织扩张,皮质破坏 **1c.** 2009 年 5 月 MR 示股骨远端髓腔积液,信号不均,外侧皮质破坏及外侧窦道 **1d.** 术前外观,切口瘢痕及愈合窦道 **1e,1f.** 2009 年 8 月 X 线片 **1g, 1h.** 2009 年 12 月 X 线片 **1i,1j.** 2010 年 3 月 X 线片示进展性的股骨远端骨溶解,皮质变薄,出现股骨远端内侧皮

质缺损 **1k.** CT 示股骨髁间窝破坏,髁间窝扩大 **1l.** 股直肌和股外侧肌下一较大“囊肿”,有包膜包裹,内容物呈豆腐渣样,大小为 10 cm×5 cm×5 cm **1m.** 完整切除囊肿后见骨缺损严重,累及股骨内外髁、髁间窝后缘、股骨干骺端、部分骨干,接近周径 1/2 **1n,1o.** 术后病理切片示表皮样囊肿伴鳞状上皮乳头瘤样增生,周围见大量中性粒细胞及巨噬细胞浸润

**Fig.1** Male, 60-year-old, chronic osteomyelitis **1a.** X-ray of January 2009 showed ill-defined patchy sclerosis and radiolucent line with periosteal reaction **1b.** CT demonstrated cortical destruction and soft tissue extension **1c.** MR image of May 2009, after the first and before the second Lautenbach procedure, showed a fluid-filled cavity with enhancing rim in the distal femoral medullary canal, with a persisting sinus tract laterally **1d.** Preoperative appearance, lateral sinus of femur **1e, 1f.** After the second Lautenbach procedure and antibiotic cement removed, bone defect along the lateral aspect from the previous Lautenbach procedure could be seen on X-rays of August 2009 **1g, 1h.** X-rays of December 2009 **1i, 1j.** X-rays of March 2010 showed progressively increasing osteolysis, marked expansion and endosteal thinning **1k.** CT showed intercondylar fossa was damage **1l.** A cyst was seen below the rectus femoris and packaged by an envelope, Grossly, a cystic lesion measuring 10 cm×5 cm×5 cm and containing white cheese-like material **1m.** The scope of the bone defects was close to half of the perimeter and overlapped with he medial and lateral femoral condyle, the intercondylar fossa, the femoral metaphysis, and part of the diaphysis **1n, 1o.** Histopathologic appearance, the cyst wall was lined by squamous epithelium with keratinizing cellula rdebris, surrounding the cyst showing giant cell reaction, consistent with the rupture of the cyst and secondary granulomatous inflammation

髓腔积液,信号不均,外侧皮质破坏及外侧窦道(图 1c)。行病灶清除加抗生素骨水泥填充术,炎症症状消失,ESR 和 CRP 正常。2009 年 8 月后予以行骨水泥去除术和外侧缺损处股外侧肌瓣填充,肿痛消失。2010 年 3 月患者左大腿肿胀疼痛,行走后加重,伴有左膝屈伸不利,窦道时有白色粉末样分泌物流出,为进一步诊治来我院就诊。否认外伤史和各系统重大基础病史。查体:生命体征正常,全身浅表淋巴结未扪及,心、肺、腹未见异常。专科检查:左大腿外侧切口、窦道瘢痕(图 1d),无渗液。左膝部肿胀、压痛明显,局部皮温增高,可触及波动感,浮髌试验阳性,被动活动,屈伸活动尚可。术前膝关节穿刺液培养 2 次阴性。对比 2009 年 8 月至 2010 年 3 月 X 线片,发现进展性股骨远端骨溶解,皮质变薄(图 1e-1j);CT 示股骨髁间窝破坏(图 1k)。

手术治疗:硬膜外麻醉下行左膝、左股骨脓肿病灶清除术,术中见股直肌和股外侧肌下一较大“囊肿”,有包膜包裹,内容物呈豆腐渣样,囊肿侵及周围骨组织,骨质破坏严重,部分包膜穿透双层骨皮质达股骨髁间后缘,包膜完整,大小 10 cm×5 cm×5 cm(图 1l)。破坏骨质边缘硬化。膝关节股骨端可见炎性滑膜形成,部分侵及内侧半月板。术中予以完整清除囊肿病灶,切除包膜和部分炎性滑膜,因骨缺损范围较大,累及股骨内外髁、髁间窝、股骨干骺端、部分骨干,接近周径 1/2(图 1m)。术后石膏固定防止骨折,考虑 II 期行腓骨移植术修复骨缺损。囊肿病灶物送病理,病理诊断为:“左股骨远端”表皮样囊肿伴鳞状上皮乳头瘤样增生。免疫组化:P53(-),Ki-67 30%(+),CK 广谱(+),EGFR(-)(病理切片见图 1n, 1o)。出院后支具保护下能下地行走,2 个月后复查患膝肿痛,摄 X 线片示左股骨远端病理性骨折。拟行人工关节置换术,后应患者要求行截肢术。

## 讨论

骨内表皮样囊肿(intraosseous epidermoid inclusion cyst)是罕见的良性骨病损,以颅骨和远端指趾骨多见,发生于长管状骨股骨远端的表皮样囊肿十分罕见。发生于长管状骨的表皮样囊肿国内外报道仅 5 例,国外报道胫骨 2 例、尺骨 2 例<sup>[1-4]</sup>,国内报道胫骨 1 例<sup>[5]</sup>,未见有巨大股骨远端表皮样囊肿的报道。根据表皮样囊肿起源可分先天性和植入性两种,后者是由于上皮组织嵌入骨组织内生长形成,往往有外伤史<sup>[4]</sup>,或手术穿刺史<sup>[6]</sup>,外伤或穿刺后鳞状上皮岛状增生植入骨内,慢性增生最终导致骨破坏。Lalam 等<sup>[7]</sup>报道 1 例在慢性骨髓炎行病灶清除术后存在于死腔内的进展型表皮样增生,伴有骨溶解,但

进展缓慢,且无任何症状。查阅国内外文献,本例进展迅速的骨内表皮样囊肿,应为国内外首次报道。

目前慢性骨髓炎临床上广泛应用 Lautenbach 病灶清除术,包括病灶清除、死骨切除,持续闭式抗生素灌注引流及抗生素骨水泥应用,最早是由 Weber 和 Lautenbach<sup>[8]</sup>在 1986 年提出,主要用于全髋置换术后感染的翻修术中处理。最近,用该方法治疗慢性骨髓炎获得较好疗效,但该方法需大量彻底清除坏死骨和肉芽组织,故可留下较大死腔,纤维结缔组织充填髓腔困难,在扩髓过程中,表皮碎屑的植入导致了表皮样囊肿在股骨死腔内的填充。Magalhaes 等<sup>[9]</sup>通过动物实验证实了表皮碎屑的植入可在股骨形成骨内表皮样囊肿。

总之,尽管本例表皮样囊肿是继发于慢性骨髓炎的病灶清除术,但在其他可能导致死腔的骨病中亦会发生。今后,将加强类似骨内死腔的管理。

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