

- Cai H, Qiao RQ. External fixator in combination with small splint for the treatment of humeral shaft fracture[J]. Zhongguo Gu Shang/China J Orthop Trauma, 2006, 19(9):513-515. Chinese.
- [10] Goodship AE, Cunningham JL, Kenwright J. Strain rate and timing of stimulation in mechanical modulation of fracture healing[J]. Clin Orthop Relat Res, 1998, 335: 105-115.
- [11] 喻鑫罡, 张先龙, 曾炳芳. 骨折段低频可控微动影响骨痂矿化与力学特性的实验研究[J]. 上海交通大学学报: 医学版, 2008, 28(12): 1491-1495.
- Yu XG, Zhang XL, Zeng BF. Experimental study on callus calcification and mechanical property of healing bone under low frequency and controlled micromovement[J]. Shang Hai Jiao Tong Da Xue Xue Bao; Yi Xue Ban, 2008, 28(12): 1491-1495. Chinese.
- [12] Epari DR, Taylor WR, Heller MO, et al. Mechanical conditions in the initial phase of bone healing[J]. Clin Biomech (Bristol, Avon), 2006, 21(6): 646-655.
- [13] 张元民, 王志彬. 小夹板治疗骨折的微观观[J]. 中国骨伤, 2000, 13(12): 722-723.
- Zhang YM, Wang ZB. Small splint to treat fracture of micromovement view[J]. Zhongguo Gu Shang/China J Orthop Trauma, 2000, 13(12): 722-723. Chinese.
- [14] 刘振利, 顾云伍, 张会生. 小夹板固定对前臂微循环影响的实验研究[J]. 中国骨伤, 1997, 10(3): 20.
- Liu ZL, Gu YW, Zhang HS. Effect of small splint fixation on microcirculation of forearm and experimental study[J]. Zhongguo Gu Shang/China J Orthop Trauma, 1997, 10(3): 20-21. Chinese.
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• 病例报告 •

## 先天性双侧胸锁乳突肌挛缩 1 例报告

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关键词 胸锁乳突肌; 挛缩; 病例报告

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**Congenital bilateral sternocleidomastoid contracture: a case report** SHI Xiao-lei, LI Chang-sheng, ZHOU Kun-peng, YANG Chen, QI Xin, and LI Shu-qiang. Department of Orthopaedics, the First Hospital of Bethune, Jilin University, Changchun 130021, Jilin, China

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患儿, 男, 10 岁, 因头部向左偏斜 6 年, 双侧胸锁乳突肌挛缩 1 个月来我院就诊。6 年前患儿家长发现其喜欢将头部偏向左侧, 写字时明显, 家长误以为是孩子的不良习惯, 仅教育孩子改正。上小学后, 头部偏斜减轻, 但逐渐出现抬头困难, 不能向两侧完全扭头, 一直未去医院诊治。1 个月前家长无意间发现患儿双侧颈部肌肉紧张明显, 双侧面部轻度不对称, 左面部稍小。患者既往婴儿期双侧颈部曾出现过硬结, 随后逐渐消退。患儿足月顺产, 臀位产, 围产期检查未发现异常。体格检查: 双侧面部轻度不对称, 右侧较饱满, 左侧稍小, 双眼斜视不明显, 颈短, 头枕部稍向左侧偏斜, 下颏转向右侧肩部。双侧胸锁乳突肌紧张挛缩, 无压痛, 呈条索状, 左侧较重, 左侧锁骨胸骨端较右侧高约 1.0 cm, 颈部各个方向活动明显

受限, 被动活动时双侧胸锁乳突肌牵拉痛阳性 (图 1a)。实验室检查: 白细胞  $4.25 \times 10^9/L$ , 中性粒细胞百分比 53%, 中性粒细胞绝对值  $2.26 \times 10^9/L$ , 碱性磷酸酶 170 U/L, 尿素氮 8.02 mmol/L, 肌酐  $46.5 \mu\text{mol/L}$ , 尿蛋白阴性, 尿酮体阴性。颈椎正侧位 X 线片示颈椎曲度凸向右侧, 齿状突略偏向右侧, 颈椎排列序列良好, 无椎体畸形与破坏, 左侧锁骨的胸骨端较右侧高 (图 1b), 骨盆正位 X 线片示双侧髋关节发育良好, 无发育性髋关节发育不良 (图 1c)。临床诊断为先天性双侧胸锁乳突肌挛缩。术前准备完毕后, 于全麻下行左侧胸锁乳突肌松解术, 手术取左侧锁骨上 1 横指处横行切口, 切口长约 3.0 cm, 术中细心操作, 仔细分离, 同时注意保护周围血管和神经, 游离出左侧胸锁乳突肌止点, 术中见胸锁乳突肌胸骨头和锁骨头均紧张挛缩, 以锁骨头较显著。首先在肌肉止点处完全切断左侧胸锁乳突肌的锁骨头及部分胸骨头, 并切取胸锁乳突肌锁骨头止点上方 2.0 cm 处肌

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图 1 患儿,男,10 岁,先天性双侧胸锁乳突肌挛缩 1a. 体格检查示头部稍向左侧倾斜,面部轻度不对称,颈短。双侧胸锁乳突肌紧张,左侧较明显,颈部各个方向明显活动受限 1b. 术前颈椎正侧位 X 线片示颈椎曲度凸向右侧,齿状突略偏向右侧,无椎体畸形及破坏 1c. 术前骨盆正位 X 线片示髋关节发育良好,无发育性髋关节发育不良 1d. 术后病理学检查见胸锁乳突肌纤维化,玻璃样变性(HE×200) 1e. 术后第 2 天开始佩戴颈托 1 个月,将头颈部制动于中立位

**Fig.1** A 10-year-old boy with congenital bilateral sternocleidomastoid contracture 1a. Physical examination showed the boy appeared to be having a short neck with mild head tilt to the left side and minimal facial asymmetry. Both sternocleidomastoid muscles were tight, and the left side was more obvious. The sternal end of clavicle was approximately 10 cm higher on the left side, movements of neck were limited obviously in all planes 1b. Preoperative AP and lateral X-rays of the cervical vertebra showed cervical vertebra curve was convex to the right side without vertebral deformity and damage, and the odontoid was also convex to the right side slightly 1c. Preoperative AP X-ray of the pelvis showed the hip was well-developed without developmental dysplasia 1d. Pathological examination by HE staining showed there was fibrosis and hyaline degeneration of the sternocleidomastoid muscles (HE×200) 1e. Postoperative photograph showed the boy's neck was fixed in neutral position with a semirigid cervical collar for 1 month, which started 2 days postoperatively

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肉组织行病理检查,转动头部,见左侧胸锁乳突肌及颈部软组织仍较紧张,随后完全切断胸锁乳突肌胸骨头并松解周围挛缩的颈阔筋膜及血管鞘等软组织,反复转动头部,明确松解适度后,结束手术。术后病理回报:胸锁乳突肌纤维化,伴有玻璃样变性(图 1d)。术后第 2 天起佩戴颈托至术后 1 个月,将头颈部制动于中立位(图 1e)。术后 1 周开始进行增加活动范围的练习,持续到术后 3 个月。术后 3 个月复查,切口愈合优良,头、面、颈部均基本对称,未出现右侧斜颈,颈部活动范围明显增大。

## 讨论

(1) 胸锁乳突肌挛缩的概述。单侧胸锁乳突肌挛缩引起的肌性斜颈及其继发性的畸形改变(如面部不对称、斜头畸形以及颈椎侧凸)非常常见<sup>[1]</sup>,其发病率大约为 0.4%~1.9%<sup>[2]</sup>。但是双侧胸锁乳突肌同时挛缩却几乎是闻所未闻的<sup>[3]</sup>。查阅国内外相关文献发现,到目前为止有关双侧胸锁乳突肌同时发生挛缩的病例文献中仅有 2 例报道,1 例是北京儿童医院贾和庚<sup>[4]</sup>在 1986 年报道的 13 岁男性患儿,

另 1 例是尼泊尔 Babu 等<sup>[3]</sup>在 2009 年报道的 19 岁的女性青少年患者,当时在对英文文献的回顾中发现,双侧胸锁乳突肌挛缩尚未有报道。

(2) 胸锁乳突肌挛缩的病因。单侧胸锁乳突肌挛缩的确切病因尚不清楚,目前有以下几种假说,如分娩时颈部软组织受压造成静脉闭塞<sup>[5]</sup>,子宫内拥挤<sup>[6]</sup>及神经性原因<sup>[7]</sup>等。目前多认为臀位产、产伤及牵拉等因素导致胸锁乳突肌损伤出血,血肿机化、挛缩形成。此外,此症有 1/5 的患儿有明确的家族史,且此类患儿常合并先天性髋臼发育不良等其他部位畸形,故认为其发生同遗传有关。最新的理论指出在胸锁乳突肌内存在来源于胚胎发育期间的间充质细胞<sup>[8]</sup>。以上这些理论是否同时也适合于双侧胸锁乳突肌挛缩尚未可知,有待进一步研究。本例患儿出生时为臀位产,考虑其发病可能与产伤有关,分娩时臀先露,胎头娩出困难,颈部软组织受压造成两侧胸锁乳突肌损伤、出血,随后血肿机化、挛缩。

(3) 胸锁乳突肌挛缩的诊断和鉴别诊断。诊断主要根据临床表现和颈椎 X 线片,患者双侧胸锁乳突

肌均紧张并挛缩,呈条索状,颈短,头部各个方向活动明显受限,颈椎 X 线片排除颈椎骨质发育畸形,即可明确诊断。需与本病鉴别的疾病有先天性肌斜颈和骨性斜颈。先天性肌斜颈是单侧胸锁乳突肌纤维性挛缩,导致颈部和头部向患侧偏斜畸形,严重者导致颈椎侧凸畸形。双侧胸锁乳突肌挛缩是双侧胸锁乳突肌均有不同程度的纤维性挛缩,若双侧挛缩程度不对称,亦可出现头颈部轻度向挛缩重的一侧偏斜,但面部不对称及斜视不明显,颈短。二者的颈椎 X 线片除了可能有轻微的颈椎侧凸外,无其他异常,这一点可与骨性斜颈相鉴别。骨性斜颈是颈椎骨质发育畸形所致的斜颈,较少见,包括短颈畸形、颅底凹陷、半椎体畸形、寰枢椎半脱位及齿状突发育畸形,这些疾病可造成斜颈及面部不对称,但一般不会产生胸锁乳突肌的典型条索状挛缩带及肿块,X 线检查可鉴别。

(4)胸锁乳突肌挛缩的治疗方法。单侧胸锁乳突肌挛缩在婴儿期可采取保守治疗,即按摩、热敷和手法被动牵拉等。保守治疗失败或者是发现较晚的年长儿则可行手术治疗。原则上是年龄越小疗效越好,因为年龄越小的患儿,胸锁乳突肌粘连越轻;反之,年龄越大粘连越重,术后畸形纠正亦较慢。Chandler 等<sup>[9]</sup>也提倡早期手术,但是目前大多数学者认为在患儿 1~4 岁行手术治疗亦能取得良好的手术效果。年龄较大的儿童斜颈手术效果欠佳,因为年长儿对斜颈相关的面部不对称等畸形的修复重塑潜能已明显降低,以及胸锁乳突肌周围软组织广泛纤维化。手术治疗方法包括胸锁乳突肌单侧头切断术、双侧头切断术、胸锁乳突肌“Z”字延长术、胸锁乳突肌全切除术及微创手术等。Cheselden<sup>[10]</sup>首次描述了胸锁乳突肌起点肌腱切断术。Lee 等<sup>[2]</sup>曾采用胸锁乳突肌起点肌腱切断术治疗青少年胸锁乳突肌挛缩取得了良好的效果。贾和庚<sup>[4]</sup>和 Babu 等<sup>[3]</sup>治疗上也采用双侧的胸锁乳突肌起始点肌腱切断术,术后 3 个月及半年随访时发现,颈部各个方向活动范围明显增大,头部倾斜,面部不对称、短颈等畸形均消失,术后效果令人满意。本例采用了相似的手术方法,但不同的是,本次手术仅在挛缩较重的一侧进行胸锁乳突肌起始点切断术,术中完全切断左侧胸锁乳突肌的锁骨头和胸骨头,并适度松解周围挛缩的颈阔筋膜及血管鞘,术后在功能和美观方面都取得了良好的效果。笔者只进行左侧胸锁乳突肌松解的理由有以下几点:①患儿双侧胸锁乳突肌均挛缩,左侧较重,表现为头部轻度向左侧倾斜,笔者试图进行较重一侧胸锁乳突肌适度松解来调节双侧颈部肌肉的力量平衡,如果术后效果满意就避免了右侧手术,大大减

少了手术创伤;如果术后出现头向右侧倾斜,可二次行右侧胸锁乳突肌松解术,也不影响手术的整体效果。②患儿双侧胸锁乳突肌均挛缩较重,若双侧同时行手术切断,术后有气管塌陷导致呼吸困难的可能性,甚至有造成呼吸衰竭的危险。③同时行双侧手术治疗,需行双切口,由于 2 个切口距离较近,切口间皮肤血供较差,术后可能出现切口间皮肤坏死。

总之,由于双侧胸锁乳突肌同时发生不同程度的挛缩,头部歪斜、面颈部不对称等发育性畸形均不明显,因此该病很容易被家长和医生忽视,导致漏诊和误诊。当患儿颈部活动范围明显减小,或头部轻度歪斜及面部发育不对称时,才会引起家长的重视,从而延误诊断和最佳治疗时机。因此在诊治该病时,临床医生应详细询问病史和进行细致的体格检查,并结合辅助检查排除颈部活动范围减小的其他原因,比如融合椎等骨性异常,做到早期诊断、早期治疗。总之,先天性双侧胸锁乳突肌挛缩非常少见,有关其诊断及治疗方面的经验尚匮乏,有待在临床工作中进一步总结。

#### 参考文献

- [1] Canale ST, Griffin DW, Hubbard CN. Congenital muscular torticollis; a long-term follow-up[J]. J Bone Joint Surg Am, 1982, 64(6): 810-816.
- [2] Lee EH, Kang YK, Bose K. Surgical correction of muscular torticollis in the older child[J]. J Pediatr Orthop, 1986, 6(5): 585-589.
- [3] Babu MK, Lee P, Mahadev A. Congenital bilateral sternocleidomastoid contracture; a case report[J]. J Pediatr Orthop B, 2009, 18(3): 145-147.
- [4] 贾和庚. 先天性双侧胸锁乳突肌挛缩 1 例报告[J]. 中华小儿外科杂志, 1986, 7(6): 344.  
Jia HG. Congenital bilateral sternocleidomastoid contracture; a case report[J]. Zhonghua Xiao Er Wai Ke Za Zhi, 1986, 7(6): 344. Chinese.
- [5] David JR, Wenger DR, Mubarak SJ. Congenital muscular torticollis; sequela of intrauterine or perinatal compartment syndrome[J]. J Pediatr Orthop, 1993, 13: 141-147.
- [6] Ling CM. The influence of age on the results of open sternomastoid tenotomy in muscular torticollis[J]. Clin Orthop Relat Res, 1976, (116): 142-148.
- [7] Sarnat HB, Morrissy RT. Idiopathic torticollis; sternocleidomastoid myopathy and accessory neuropathy[J]. Muscle Nerve, 1981, 4(5): 374-380.
- [8] Tang S, Liu Z, Quan X, et al. Sternocleidomastoid pseudotumour of infants and congenital muscular torticollis; fine structure research [J]. J Pediatr Orthop, 1998, 18(2): 214-218.
- [9] Chandler FA, Altenberg A. Congenital muscular torticollis in children[J]. JAMA, 1944, 125: 476-483.
- [10] Cheselden W. Torticollis. In: LeDran FW, editor. The operations in surgery (trans. T Gataker)[M]. London: C Hitch and R Dodsley, 1949: 454.

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