

先天性食管闭锁并气管食管瘘的诊断与治疗现状

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收稿日期: 2016-04-29
修回日期: 2016-05-24
接受日期: 2016-06-06
在线出版日期: 2016-12-08

Diagnosis and treatment of congenital esophageal atresia with tracheoesophageal fistula

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Received: 2016-04-29
Revised: 2016-05-24
Accepted: 2016-06-06
Published online: 2016-12-08

Abstract

Esophageal atresia with or without tracheoesophageal fistula (EA/TEF) is a congenital life-

threatening malformation which requires surgical repair, but it is still a challenge for patients and surgeons because of EA itself, possible combined severe deformities, and surgical risk. Thanks to the development and improvement of diagnostic and therapeutic methods and techniques, especially the progress achieved in preoperative EA diagnosis, successful surgery for long-gap EA/TEF, and the application of thoracoscopic technology, the survival rate after surgery has reached 95%. However, the possible postoperative complications and its managements should not be ignored.

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Key Words: Esophageal atresia; Tracheoesophageal fistula; Diagnosis; Treatment; Complications

Wu XD. Diagnosis and treatment of congenital esophageal atresia with tracheoesophageal fistula. *Shijie Huaren Xiaohua Zazhi* 2016; 24(34): 4537-4541
URL: <http://www.wjgnet.com/1009-3079/full/v24/i34/4537.htm> DOI: <http://dx.doi.org/10.11569/wcjd.v24.i34.4537>

摘要

食管闭锁并气管食管瘘(esophageal atresia with or without tracheoesophageal fistula, EA/TEF)是一种需要手术修复的威胁生命的先天畸形, 但不论是食管闭锁本身、可能存在的严重合并畸形还是手术的实施, 仍是对患儿和医者的考验和挑战. 随着诊疗方法和技术的发展与提高, 尤其是术前对EA的分型诊断、长段缺失型手术成功的积累增多及胸腔镜技术的应用等均取得进展, 手术存活率已达到95%, 但术后并发症及其处置仍需

■背景资料

随着诊疗方法和技术的发展与提高, 食管延期一期吻合术、食管替代术和胸腔镜技术等的应用不仅使长段型先天性食管闭锁(esophageal atresia, EA)得以治疗, 而且手术存活率达到了较高的水平, 但在积累越来越多长段型EA/气管食管瘘(tracheoesophageal fistula, TEF)成功修复经验的同时, 与之有关的术后并发症也增多, 因此, 必须重视长段型根治手术的选择和术后并发症的处置.

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研究前沿

EA的分型基于是否存在TEF, 但EA段长短与手术方式选择和手术效果关系密切, 尤其以长段缺失型手术难度大且并发症多。因此, 本文聚焦术前分型诊断和对EA段的测量及长段型EA根治手术的决策。

不断得到重视。

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关键词: 食管闭锁; 气管食管瘘; 诊断; 治疗; 并发症

核心提要: 食管闭锁(esophageal atresia, EA)是一种需要手术修复的威胁生命的先天畸形, 尤其长段食管缺失的EA往往需要多次手术且修复难度大、并发症多, 手术前明确闭锁类型和闭锁段长短对决策根治手术方式和减少并发症有重要意义。

吴学东. 先天性食管闭锁并气管食管瘘的诊断与治疗现状. 世界华人消化杂志 2016; 24(34): 4537-4541 URL: <http://www.wjgnet.com/1009-3079/full/v24/i34/4537.htm> DOI: <http://dx.doi.org/10.11569/wjcd.v24.i34.4537>

0 引言

先天性食管闭锁(esophageal atresia, EA)并不罕见, 在出生活婴中的发生率为1/(1500-5000)^[1,2], 可单独存在, 但多同时存在闭锁远端或近端气管食管瘘(tracheoesophageal fistula, TEF), 即EA/TEF。这是一种威胁生命的畸形, 不仅因为EA/TEF本身及其可能的合并畸形, 也因为手术介入和可能发生的致命并发症, 所以, 手术治疗该病不仅是对医者的考验, 更是对生命的挑战, 在过去的60年里, 手术存活率逐渐提高, 现已超过90%, 部分发达地区已超过95%^[3-5], 而时至今日能否实施EA/TEF手术仍然是小儿外科实力和技术水平的标志。EA/TEF的诊断已多无问题但对术前分型的认识上仍有差异, 而对不同类型的EA/TEF手术经验也各异, 尤其是对长段缺失型闭锁, 术后转归尤其是远期随访结果尚有待完善。

1 进行术前EA/TEF分型诊断的重要性

通常根据病史、临床表现和鼻(口)胃管造影可确立EA/TEF的诊断。对于孕中后期羊水过多、生后喂养困难和口溢泡沫等就应怀疑存在EA的可能, 发现新生儿腹部不膨隆且放置胃管受阻不能到达胃并抽得胃液时, 存在EA的可能性很大, 经胃管注射造影剂摄X片即可确诊并能初步分型。

食管腔不连续即为EA, 可合并或无TEF,

或虽食管腔连续即未闭锁但存在TEF, 通常将EA/TEF分成5型: I型即食管腔不连续, 闭锁上下端各形成盲端, 无TEF; II型即闭锁的食管上端与气管相通, 下段呈盲端; III型即食管上段为盲端而下端与气管相沟通; IV型即闭锁食管的上、下端分别与气管相沟通; V型虽无EA但存在TEF, 又称H型。其中III型最为常见, 占总数的90%左右, 另外4型仅为总数的10%左右^[6]。I型因无TEF, II型虽有瘘但吞咽的气体均不能进入胃肠道, 因而生后腹部不膨隆, 摄腹部X线平片可无气体影, 其他各型均可见腹部膨隆且腹部X线平片可见气体影。

各型EA/TEF均需手术才能矫治, 但手术前明确食管缺失段的距离是非常重要的, 便于根据缺失段的长短选择手术方式和是否分期手术, 因此又根据食管缺失段的长度分为缺失<2.0 cm并可一期吻合的短段型、缺失2.0-3.5 cm的长段型和>3.5 cm的超长段型^[7], 后二者的手术修复较为困难且治疗周期较长, 可选择延期一期吻合、二期修复或食管替代术。经鼻(口)胃管造影可明确EA/TEF的诊断和初步分型, 但难以得知食管缺失段的长度, 以往多在剖胸探查和胃造瘘的基础上同时经胃管和造瘘管注射造影剂摄X片, 可准确测量食管显影两盲端间的距离, 现在鼻(口)胃管造影的基础上通过高分辨率B超和多层螺旋CT(multislice helical CT, MSCT)三维重建及容积再现技术能显示食管缺失段的长度^[8,9], 因此, 术前有条件应作MSCT扫描, 既无创也准确。

2 EA/TEF手术的策略

随着术前EA/TEF分型诊断技术和对食管缺失段长度测量准确性的提高, 以往常规将胃造瘘术作为分期一期手术已不再常规推荐, 因为多数EA/TEF均能一期修复, 但是EA/TEF常合并其他畸形或为早产儿、低体质量儿或极低体质量儿, 或因生后不能及时作出EA/TEF的诊断并采取恰当有效的护理指导, 导致发生吸入性肺炎等, 这些因素都将使治疗的决策和实施变得更加困难。尽管随着诊疗措施的增加和技术的提高, 体质量和肺炎已不再是影响预后的重要因素, 但低体质量伴有严重心血管畸形者却是与手术成活率关系较为紧密的因素, 尤其是体质量<1500 g且合并严重的心血管畸形者, 手术存活率降低, 生存质量也较差^[10,11]。

相关报道

在对EA段的术前测量中, 苏鹏俊等和汪松等分别描述了高频超声和多层螺旋CT的应用及结果, 而郑珊等、黄金狮等和Huh等分别报道长段型EA手术治疗中延期一期吻合中内或外牵引的应用和胸腔镜技术的应用; Schneider等和Chang等报告了法国和韩国EA术后近期的随访结果, 而Kovesi等及Huynh-Trudeau等均分别报告了远期甚至到成人的随访结果; Maghsoudlou等报告了食管干细胞的研究结果。

食管本身是修复EA/TEF最好的材料^[6], 一期或延期一期吻合术应作为首选的修复方式. 对于绝大多数III型(包括IIIA和IIIB两个亚型)均可在结扎并修补TEF后作食管一期吻合术尤其是食管缺失段<1.5 cm的IIIB型, 大部分IIIA型和部分长段型可经努力后一期吻合即食管延长吻合或延期一期吻合术, 均有相应成功的经验值得借鉴, 有游离食管延长、近端食管瓣翻转、食管肌层螺旋切开延长等, 对于经游离后仍难以进行吻合者, 内牵引或外牵引1 wk后再吻合即延期一期吻合术也有成功的经验^[12-14]. III型中IIIB型多见, 缺失段短, 可直接吻合, 对吻合口的血供影响较小且吻合张力不大, 术后发生并发症较少, 生活质量较高, 但对于缺失段>1.5 cm者或经延长后的吻合仍有张力者, 发生吻合口瘘或狭窄的机会增加. 当然, 除了吻合口血供和张力外, 吻合技术是最为重要的防止吻合口瘘和狭窄的措施, 通常采用的是黏膜对黏膜的一层吻合, 进行二层吻合者, 瘘和狭窄的机会增加^[1,3,15]. 有的作者报告通过闭锁两盲端交叉“+”字切开后嵌合吻合合法对预防狭窄是有帮助的^[16].

对于超长段型难以作一期或延期一期吻合者可选择分期手术或食管替代术^[17]. 分期手术中, 一期完成胃造瘘并经肠营养, 食管上端持续牵引, 术后采取头低脚高位刺激食管生长, 加强营养支持并积极预防吸入性肺炎, 8 wk后再行食管吻合术或食管替代术^[12], 这可降低新生儿期手术的风险, 不仅手术安全而且术后并发症也相对较少; 食管替代材料可根据术者经验选择管状胃、全胃、结肠、小肠或回盲部进行替代^[18]. 由于长段型手术修复困难, 除进行延期一期吻合或自身器官替代外, 组织工程食管将可能是解决该问题的努力方向, 目前已有作者^[19]开始研究食管干细胞的分离、培养, 但仍处于探索阶段, 一旦获得成功, 将给长段型EA的修复更多选择.

另有作者报道^[20]一种特殊类型即闭锁近端食管盲端缺失并气管远端食管瘘, 但尚未看到对此类型EA修复成功的报道, 当然, 先作TEF修补和胃造瘘并进行肠内营养是挽救患儿生命的必要措施.

随着腔镜技术的发展和应用越来越广泛, 胸腔镜下食管吻合术已应用于各型EA的修复, 其中在长段型修复中的应用已凸显微创

的优势^[13,21,22].

尽管食管本身是最理想的修复EA的材料, 也有越来越多的分期或延期一期吻合成功修复长间隙EA的报道, 但长间隙EA术后狭窄的机会较多, 切除狭窄段后仍可能再狭窄^[23], 而且发生胃食管反流、吞咽困难等其他并发症的机会也增多, 因此, 针对食管缺失达到或超过6个椎体的超长段EA根治手术的选择, 除了术者的经验和可利用的便利条件外, 食管替代术可能在提高手术效果和远期生存质量上更有优势.

3 术后并发症和处置

文献报道术后并发症的发生率差异较大, 包括结构性和功能性的问题, 但吻合口瘘和狭窄仍是围手术期和1年内主要、重要而且需要进行处置的并发症, 其他并发症如胃食管反流等仅极少数需要手术介入^[3,24-28], 随着全静脉肠外营养和经空肠营养管肠内营养支持的进展^[29], 未继发胸腔感染的吻合口瘘已不再是威胁生命的并发症, 但由于长段型EA修复成功的病例积累逐渐增多, 食管延长吻合术的应用等, 术后食管狭窄的报道也增加, 其中端端吻合术后吻合口狭窄的发生率较食管胃管吻合者多^[3,6,30], 而这仍是影响生活质量并需要积极处理的问题.

食管扩张术是被普遍采用处置术后食管狭窄的方法, 该方法不仅需要频繁实施, 而且痛苦较明显, 也由于操作技巧问题, 食管穿孔、TEF复发等时有发生^[31,32], 内镜下超声和食管扩张不仅可以诊断狭窄的原因并可获得较理想的扩张效果, 而且可降低食管穿孔的发生率^[33,34]. 以往, 对于严重瘢痕食管狭窄可选择狭窄段切除再吻合, 但仍难以避免再度狭窄^[23], 为此有作者报道^[2]植入食管支架可使食管得以持续扩张并缓解吞咽困难的状况.

尽管吻合口瘘和狭窄仍是EA术后常见的并发症, 但由于越来越多的长段型EA得以治疗, 术后并发症的发生率也增加, 有的甚至达到100%, 尤以超长段缺失型为著^[7], 而有的并发症是在患者生长发育过程中逐渐显现, 虽然多数均可通过保守治疗而长期缓解, 有的并发症却长期困扰患者的生活, 导致营养不良、生长发育迟滞等^[35-38], 有的甚至需手术如采用Nissen手术治疗返流等, 因此, EA术后患者需

□创新盘点

本文作为二次文献, 聚焦目前已报道的关于EA术前分型诊断, 对EA段的测量、长段型EA根治手术方式及手术并发症等的原始报道, 针对长段型EA的诊断和治疗方法进行评价, 为长段型EA的手术决策提供了依据.

□应用要点

随着长段缺失型EA手术成功的积累增多, 相关的并发症也随之增加, 本文的撰写对如何更合理的选择长段型的根治手术和减少并发症具有积极意义, 尤其倡导对超长段缺失型EA根治手术的决策需要循证化.

□名词解释

延期一期吻合术: 是指在实施食管吻合术前或术中确定为长段型或经游离后闭锁食管两盲端间仍有一定距离难以完成端吻合时, 先作内牵引或外牵引, 1 wk后再实施的食管吻合术。

长期甚至终生随访。

4 结论

随着诊疗方法的发展和技术的提高, 术前可对EA/TEF作出明确的分型诊断并准确测量食管缺失段的长度, 为长段型EA的手术决策提供了依据, 食管吻合术、食管替代术及胸腔镜技术等的应用不仅使长段型EA得以治疗, 而且手术存活率达到了较高的水平, 但在积累越来越多长段型EA/TEF成功修复经验的同时, 需重视与之有关的术后并发症, 对超长段缺失型EA根治手术的决策也需循证化。

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同符评价
本文对EA/TEF的分型诊断, 食管缺失段长度测量在治疗中的应用及术后并发症的处理进行系统综述, 对EA/TEF的临床诊疗具有一定指导意义。

编辑: 郭鹏 电编: 胡珊





Published by **Baishideng Publishing Group Inc**
8226 Regency Drive, Pleasanton,
CA 94588, USA
Fax: +1-925-223-8242
Telephone: +1-925-223-8243
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ISSN 1009-3079

