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·病例研究·

先天性泪腺皮肤瘘伴异位泪腺：病例报告与文献回顾

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文章亮点

1. 关键发现

- 通过查阅文献尚未发现先天性泪腺皮肤瘘伴异位泪腺。

2. 已知与发现

- 先天性泪腺皮肤瘘伴异位泪腺临床上罕见，极易漏诊和误诊。
- 对于先天性泪腺皮肤瘘伴异位泪腺患者需谨慎设计手术方案，避免术后并发症。

3. 意义与改变

- 本文报道了一例先天性泪腺皮肤瘘伴异位泪腺患者，经过术前一系列检查，行泪腺瘘管上段转位结膜囊吻合代泪腺导管术，手术方式别有洞天，希望通过该报道，有助于加深临床医生对此类疾病的认识，为今后临床诊疗提供思路。

[摘　要]　**目的：**分析先天性泪腺皮肤瘘伴异位泪腺的临床表现，总结其治疗方案。**方法：**采用病例研究与文献回顾方法，记录1例确诊为先天性泪腺皮肤瘘伴异位泪腺患儿的外观照片、泪道探查冲洗情况、泪腺瘘管数字减影检查、泪腺瘘管CT造影检查等结果。予患者行泪腺瘘管下段及异位泪腺切除+泪腺瘘管上段转位结膜囊吻合代泪腺导管术，术中切除的病变组织行病理检查，术后随访。以“泪腺瘘”、“异位泪腺”为检索词，在 PubMed、CNKI 数据库中进行文献检索，检索到相关文献共 25 篇。**结果：**患儿为男性，5岁，眼科检查于左眼上睑中外1/3处见直径约1 mm瘘口，瘘口皮肤凹陷并有簇状毛发生长，有透明液体从瘘口阵发性流出。泪器检查示左上睑皮肤瘘管开口朝外上方，瘘管探查从瘘口进针，瘘管先是内下方走行再向外下走行，在距皮面3.5 mm处有一软性抵抗，加压不能突破，冲洗液原路反流，无脓液或血性液体反流。左眼泪腺瘘管数字减影检查示左眼泪腺瘘管造影剂存留，瘘管深部存在扇形腔隙。泪腺瘘管CT造影结果显示左眼外侧泪腺高密度影。术后随访6个月，患儿左眼上睑切口愈合好，未见瘘管复发，转位的泪腺瘘管成功将泪液引流入上穹隆结膜囊内。**结论：**先天性泪腺瘘同时合并先天性泪腺异位和毛发异位者临床上罕见，术前详细检查和精确诊断对指导治疗很有帮助；在明确主泪腺是正常的情况下，完整切除异位的泪腺组织，并行泪腺瘘管转位吻合于上穹隆结膜囊代泪腺导管术，是一种较好的治疗选择。

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电子全文

[关键词] 先天性泪腺皮肤瘘；先天异位泪腺；手术治疗；泪腺瘘管结膜吻合术
[中图分类号] R777.2+1，R779.7，R779.62 [文献标志码] A

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Congenital lacrimal gland cutaneous fistula with ectopic lacrimal gland: case report and literature review

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Abstract **Objective:** To demonstrate the clinical characteristics and surgical effects of congenital lacrimal gland cutaneous fistula with ectopic lacrimal gland. **Methods:** Observational case study and literature review. The results of appearance photographs, lacrimal duct probing and irrigation, digital subtraction imaging of the lacrimal fistula, and CT scanning of lacrimal fistula in a patient diagnosed with lacrimal gland cutaneous fistula with ectopic lacrimal gland were recorded. The surgical treatment for the patient was explored. The surgical strategy entailed resecting the lower segment of the lacrimal gland fistula and ectopic lacrimal gland, combined with transposing the upper segment of lacrimal gland fistula for conjunctival sac anastomosis, instead of using lacrimal gland catheter. Pathological examination and postoperative follow-up were conducted. **Results:** Ophthalmic examination revealed a fistula with a diameter of approximatedly 1mm in medial-temporal 1/3 of the upper eyelid of the left eye. The skin surrounding the fistula was sunken and covered with tufts of hair. There was a paroxysmal discharge of clear fluid from the fistula. Lacrimal examination showed that the opening of the fistula on the skin of the left upper eyelid was directed outward and upward. Fistula exploration was conducted through the opening. The fistula tract initially coursed medially and caudally, then laterally and caudally. At a depth of 3.5mm from the dermal surface, a soft resistance was encountered that could not be overcome with forced pressure. The irrigation fluid refluxed along its original pathway, with no pus or bloody fluid regurgination. Digital subtraction imaging of the lacrimal fistula in the left eye demonstrated that contrast media remained and formed fan-shaped spaces in the depth of the fistula. CT results of lacrimal fistula revealed a high density of lacrimal gland in left eye. During outpatient follow-up six months after surgery, the incision on the left upper eyelid was observed to have healed well, with no recurrence of the fistula The transposed lacrimal fistula successfully diverted tears into the conjunctival sac of the upper fornix. **Conclusions:** Congenital lacrimal gland cutaneous fistula combined with congenital ectopic lacrimal gland and ectopic hair is rare in clinical practice. Detailed preoperative examination and accurate diagnosis are extremely beneficial for guiding treatment. When the main lacrimal gland is normal, a better treatment choice involves completely removing the ectopic lacrimal gland tissue and transferring the lacrimal gland fistula to the conjunctival sac of the upper fornix to replace the lacrimal gland catheter.

Keywords congenital lacrimal gland cutaneous fistula; congenital ectopic lacrimal gland; surgical treatment; Lacrimal gland fistula conjunctival anastomosis

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先天性泪腺瘘管是一种泪腺导管的发育异常，发病率为万分之一至万分之四，常发生于颜面部形成时期[1]。泪腺瘘管多见于外侧上睑板的上方、外侧部和上下睑外侧部，一般不超过眶缘。泪腺瘘管的症状和体征包括泪腺瘘管口周围长有细微的毛发丛，主泪腺分泌的泪液可由皮肤开口流出[2]。症状较轻时排出的泪液可蒸发消失，症状严重时泪液排出较多，瘘管口周围会有皮肤的糜烂、湿疹，表皮剥脱，若继发感染，可出现瘘口流脓及局部皮肤红肿痛症状[3-4]。目前先天性泪腺瘘的发病机制尚未明确，可能是由胚胎发育异常、遗传因素等多种因素作用的结果[5]。异位泪腺又称为泪腺迷离瘤，是一种良性病变，指泪腺组织出现在其正常解剖位置以外的地方[6]，发病率仅为十万分之一[7]。

本文报道1例确诊为先天性泪腺皮肤瘘伴异位泪腺的患儿，表现为自幼左眼上睑皮肤小孔洞流液，在手术探查中发现异位的毛发病灶旁有异位泪腺组织，完整切除异位的泪腺组织和异位的毛发病灶，并行泪腺瘘管上段转位结膜囊吻合代泪腺导管术。术后眼睑皮肤和结膜切口愈合好，转位的泪腺瘘管成功地将泪液引流入上穹隆结膜囊内，现报告如下。

1　病例资料

患儿为男性，5岁，因“自幼左眼上睑皮肤小孔洞流液5年余”于2022-09-05到解放军总医院第三医学中心眼科医学部就诊。患儿出生后由家长发现左眼上睑皮肤有一小孔洞伴间断流清液，哭闹及长时间用眼症状加重，伴局部毛发生长。先后多次就诊于外院，诊断为“泪腺瘘”，建议转我院进一步治疗，遂到我院就诊。门诊以“左眼先天性泪腺皮肤瘘”收入院，拟手术治疗。患儿既往无手术史、外伤史、输血史，无药物、食物过敏史，预防接种史不详。足月顺产，出生体质量为3 500 g，其母曾于孕期进行保胎治疗，孕期健康，未服用特殊药物。父母健在，均体健，家中独子，家族中无传染病及遗传病史。眼科检查：视力右眼0.8+，左眼1.0。左眼上睑中外1/3处见一直径约1 mm瘘口，瘘口皮肤凹陷并簇状毛发生长(图1)，并有透明液体阵发性流出。泪器检查：双眼泪道冲洗，氯霉素尝味试验阳性；左上睑皮肤瘘管开口朝外上方，瘘管探查从瘘口进针，瘘管先是内下方走行再向外下走行，在距皮面3.5 mm处有一软性抵抗，加压不能突破，冲洗液原路反流，无脓液或血性液体反流。

初步诊断：1)左眼先天性泪腺皮肤瘘；2)左眼上睑毛发异位。患儿病情复杂，为明确诊断，为患儿行在全身麻醉下行左眼泪腺瘘管数字减影检查，结果显示左眼泪腺瘘管造影剂存留，瘘管深部存在扇形腔隙(图2)。并行泪腺瘘管CT造影检查，行眼眶CT水平+冠状位扫描，泪腺瘘管CT造影结果显示左眼外侧泪腺高密度影(图3)。后予患儿全身麻醉下行左眼泪腺皮肤瘘管探查+异位毛发病灶切除+泪腺瘘管转位结膜囊吻合代泪腺导管术。患儿取仰卧位，全身麻醉起效后，常规用75%酒精消毒左眼周围皮肤，铺无菌手术巾。自泪腺瘘管皮肤面注入美蓝标记瘘管，瘘管内插入冲洗针，翻转眼睑，暴露穹隆结膜，冲洗针引导下横行切开穹隆结膜3 mm。沿左上睑瘘口周围做梭形皮肤切口，钝性分离瘘管周围组织，术中发现瘘管基底部呈扇形，内表层为上皮组织，见毛囊及较多毛发，且有



图 1　A左眼外观照，左眼上睑中外1/3处见直径约1 mm瘘口，瘘口皮肤凹陷并簇状毛发生长，B为同一患儿的病变部位局部放大图。

Figure 1　A The left eye. A fistula with a diameter of about 1mm was observed in 1/3 of the upper eyelid of the left eye. The skin of the fistula was sunken and tufted with hair growth. B Partial enlarged detail of the diseased region.

一大小约为3 mm × 3 mm × 2 mm的异位泪腺组织，其与瘘管相连，瘘管的另一端与正常泪腺导管相连。完整切除皮肤到异位泪腺之间的瘘管和异位泪腺及异位毛发病灶，切除的病变组织(图4)送病理检查。将正常泪腺导管用8/0可吸收线固定缝合于颞上穹隆结膜，导管内置引流条1根，引流条以8-0线固定缝合于睑结膜表面，6-0丝线间断缝合皮肤切口。术毕，予患儿左眼戴角膜绷带镜，绷带加压包扎左眼。手术顺利，切口愈合好，术后2 d打开加压包扎(图6)观察，患儿左眼上睑皮肤无明显红肿，无渗血、渗液，切口对合好，缝线在位，泪腺导管引流条在位，术后 7 d 拆除缝线，取出泪腺导管引流条。术后病理报告确诊左眼先天性泪腺皮肤瘘管伴毛发异位，异位泪腺(图5)。患儿于2022年9月22日出院。术后1个月返院复查，结膜吻合口和皮肤切口愈合好(图7)，转位的泪腺瘘管成功将泪液引流入上穹隆结膜囊内。术后随访1年，未见瘘管复发。



图 2 左眼泪腺瘘管数字减影检查

Figure 2 Digital subtraction imaging of the lacrimal fistula in the left eye

左眼泪腺瘘管造影剂存留，瘘管深部存在扇形腔隙(黄色箭头)。

It showed that contrast media remained and there were fan-shaped spaces in the depth of the fistula(yellow arrow).

2 讨论

先天性泪腺瘘和先天性泪腺异位在临床上均少见，先天性泪腺瘘同时合并先天性泪腺异位和毛发异位者临床上罕见，至今未见文献报道。关于泪腺及泪腺导管的发育，在胚胎第5周，泪腺开始在眼窝外侧形成，泪腺导管开始在泪腺内形成。在胚胎第7周，泪腺开始分泌泪液，泪腺导管开始连接到结膜囊。先天性泪腺瘘的发病率为万分之一到万分之四，异位泪腺的发病率仅为十万分之一[1, 7]。异位泪腺最常见于结膜，其次为眼眶、眼睑、眼球内，甚至极少数可以见于泪囊区及鼻窦[8]。异位泪腺的诊断较为困难，因为缺乏特异性表现，术前诊断通常依赖于手术切除或活组织检查(活检)病理证实[9-11]。治疗方法多为手术切

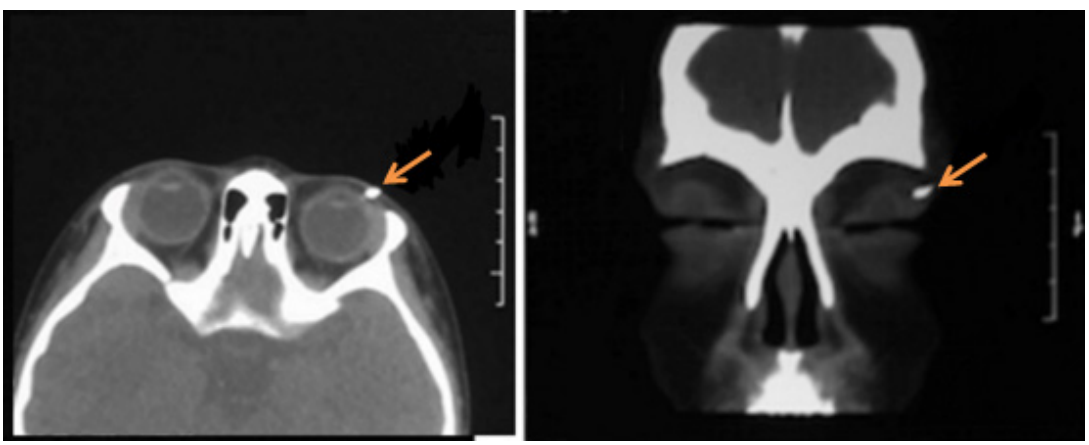


图 3 泪腺瘘管CT造影结果

Figure 3 CT results of lacrimal fistula

左眼泪腺高密度影(橙色箭头)。

It showed high density of lacrimal gland in left eye(orange arrow).



图 4　术中切除的泪腺瘘管、异位泪腺及异位毛发

Figure 4　Intraoperative removal of lacrimal fistula, ectopic lacrimal gland and ectopic hair

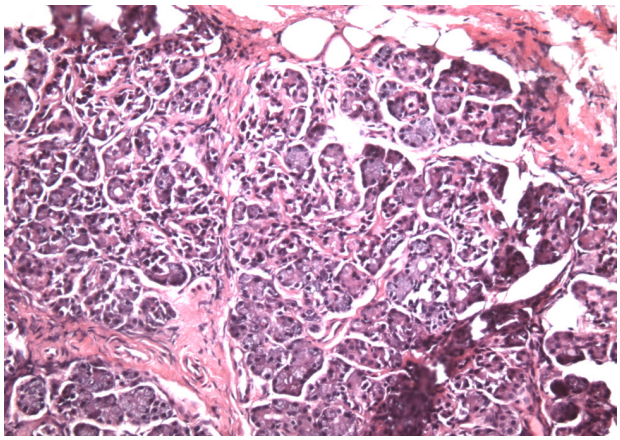


图 5　异位泪腺组织的病理图片(HE 染色，X40)

Figure 5　Pathological images of ectopic lacrimal gland tissue (HE staining, X40)

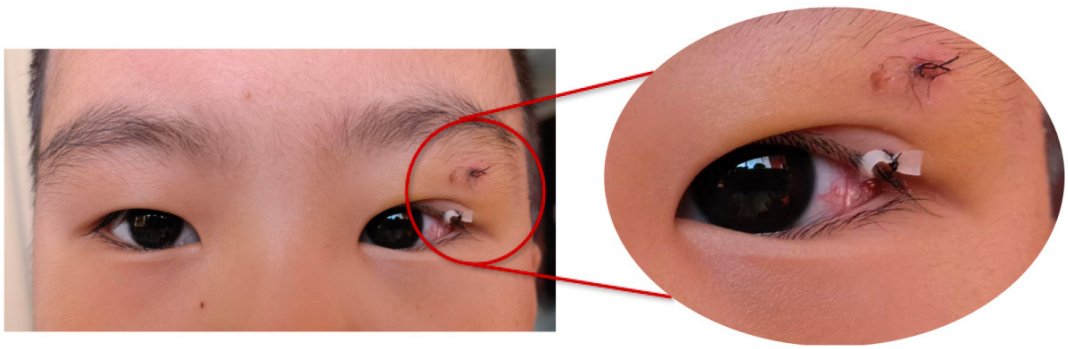


图 6　患儿术后第二天患眼外观

Figure 6　Appearance of the affected eye on the second day after surgery

术后第二天打开左眼加压包扎，切口对合好，缝线在位。

On the second day after surgery, the left eye was opened for pressure dressing. The incision was closed, and the suture was in place.

除，对于无症状的患者也可以采取保守治疗和定期复查监测[12-14]。

正常情况下，婴儿在出生时，泪腺已经发育成熟[15]。泪腺的发育主要受遗传、环境、激素和营养因素所影响[16]。本例患者异位泪腺发生在上眼睑内的泪腺皮肤瘘管旁，实属罕见。患儿瘘管位于左眼上睑中外部，瘘管开口周围伴有簇状毛发，情绪激动及久视物后清液流出加重，明显影响患儿外观和身心健康，故主动要求手术治疗。术前术者对手术方式进行了讨论：如单纯切除瘘管，存在瘘管切除不完全和后期形成泪腺囊肿和新的泪腺瘘口的可能；如果切除泪腺瘘管，并切除与之相连的泪腺小叶组织，损伤较大，容易造成泪腺导管损伤、术后泪液分泌不足的风险[17-20]；若行瘘管转位至上穹隆结膜囊，完成瘘管结膜囊吻合术，必要时放置引流条引流泪液至结膜囊，那么可以另辟蹊径，达到变废为宝、一举两得的效果。术中，术者钝性分离瘘管周围组织，暴露瘘管基底部，呈扇形，内面为上皮组织，见毛囊及较多毛发，随后在手术显微镜下发现有一疑似异位泪腺的组织团块与瘘管相连，瘘管的另一端与正常泪腺导管相连，术中完整切除瘘管及疑似异位泪腺。术式名称：左眼泪腺瘘管下段和毛发病灶及可疑异位泪腺组织切除+泪腺瘘管上段转位结膜囊吻合代泪腺导管术，泪腺皮肤瘘伴异位泪腺的诊断在术后病理诊断中被

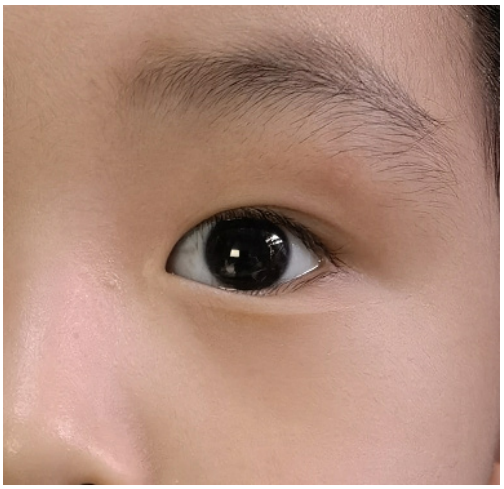


图7 患儿术后第六月患眼外观

Figure 7 Appearance of the affected eye on the sixth month after surgery

术后6个月门诊随访，患者左眼外观，上睑切口愈合好。

External observation of the left eye was observed during outpatient follow-up 6 months after surgery.

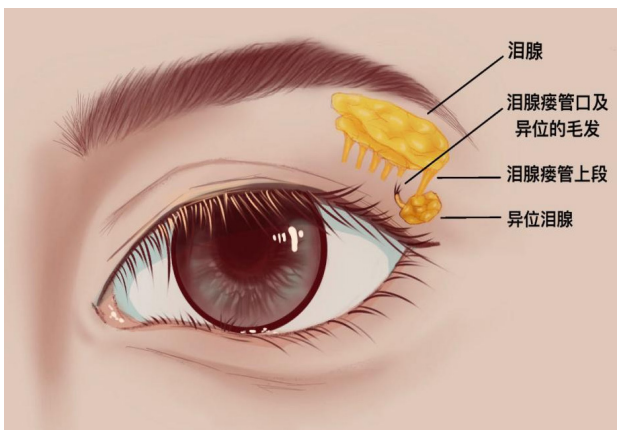


图8 左眼泪腺及瘘管位置手绘示意图

Figure 8 Schematic diagram of the position of lacrimal gland and fistula in left eye

证实。

先天性泪腺瘘需要与继发性泪腺瘘鉴别诊断。继发性泪腺瘘常发生在外伤、感染或眼睑外眦手术后；瘘管位置不固定，取决于外伤、感染、手术的部位；瘘管通常在伤口上或手术切口周围，形态不规则；同时可能会伴有泪液分泌减少或消失[20-21]。先天性泪腺瘘则是在出生时即有瘘管形成，瘘管通常较细，常位于上眼睑近外眦侧皮肤，泪液分泌通常正常[1]。

通过对本病例的诊疗过程，笔者认为值得总结的经验如下：1)对于先天性泪腺瘘和先天性异位泪腺的诊断，术前仔细行泪道瘘管探查，并行泪腺瘘管数字减影检查、泪腺瘘管CT造影等影像检查等，有利于明确病变部位和范围，指导手术设计[22-23]；2)患儿异位泪腺体积较小，术前在相应的部位并未能触及包块，术中在手术显微镜下仔细探查，在泪腺的非正常解剖位置上发现泪腺组织，是诊断异位泪腺的关键，而后期的病理检查可进一步帮助确诊[24-25]；3)对于先天性泪腺瘘伴先天异位泪腺的治疗，在明确主泪腺是正常的情况下，完整切除异位的泪腺组织，并行泪腺瘘管转位吻合于上穹隆结膜囊代泪腺导管术，是一种较好的治疗选择。

利益冲突 所有作者均声明不存在利益冲突。

开放获取声明

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