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• 防治实践 •

左前胸壁第四鳃裂畸形1例报道及文献回顾

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【摘要】 目的 探讨特殊位置第四鳃裂畸形的临床治疗要点, 为临床提供参考。**方法** 对1例发生在左前胸壁并以锁骨下方为瘘口第四鳃裂畸形患者的临床资料进行总结并结合文献分析。**结果** 该例患者主诉左侧前胸壁肿物反复肿痛2个月, 查体见左侧锁骨下方有一10 mm×10 mm瘘口, 瘘口持续流出黄色清亮的液体, 向上胸锁关节浅面颈胸交汇处触及一20 mm×20 mm×10 mm的肿物, 无波动感, 活动度欠佳, 按压稍疼痛。影像学检查显示为囊性病变。初步诊断为第四鳃裂畸形。于患者锁骨下瘘口痕迹、触及囊肿处注入少量亚甲蓝, 设计锁骨上T型切口, 翻瓣显露亚甲蓝染色的所有区域及邻近的黏膜下组织, 于胸骨上切迹颈阔肌深处找到肿块, 并沿其周围切开并切除。病理结果为第四鳃裂畸形。术后随访1周及3个月后患者均无不适, 无复发。复习相关文献表明, 第四鳃裂畸形是一种先天性发育性异常, 发生率占所有鳃裂畸形的1%, 常以瘘管、囊肿或窦道存在, 解剖位置位于颈根部、锁骨上区, 其皮肤开口靠近胸锁乳突肌的内侧下边界, 诊断时可通过其解剖位置、影像学检查结合术后病理明确诊断, 通常与颈部其他肿物如血管瘤、甲状舌管囊肿等进行鉴别诊断。手术切除是常用治疗手段, 近年文献报道经内窥镜定位及内瘘口灼烧对于多次复发的第四鳃裂畸形疗效较好, 少数可复发感染甚至癌变。**结论** 对特殊位置的第四鳃裂畸形进行治疗时, 临床医生应充分借助影像学方法明确病变的大小、解剖部位及走行, 完整、安全切除病灶, 预防病灶复发。

【关键词】 鳃裂畸形; 第四鳃裂; 鳃裂瘘; 鳃裂残余; 胸骨前区域; 胸锁关节; 囊肿

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The fourth branchial cleft deformity on the left anterior chest wall: a case report and literature review

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【Abstract】 Objective To explore the main points of clinical treatment of fourth branchial cleft deformity in special positions and to provide a reference for clinical practice. **Methods** The clinical data of one case of a fourth branchial cleft deformity that occurred in the left anterior chest wall with a fistula below the clavicle are summarized and combined with a literature review. **Results** The patient complained of repeated swelling and pain under the left anterior chest wall for 2 months. A 10 mm×10 mm fistula with yellow clear liquid exudate from the fistula was observed on the left side below the clavicle. A 20 mm×20 mm×10 mm swelling was immediately adjacent at the superficial cervicothoracic junction of the upper sternoclavicular joint, with no fluctuation and poor activity; this swelling produced slight pain upon pressing. Imaging examinations pointed to cystic lesions. The primary diagnosis was a fourth branchial deformity. A small amount of methylene blue was injected into the patient's subclavian fistula, and a supraclavicular T-shaped incision was made where the cyst contacted the fistula. By turning the flap, all the methylene blue-stained areas

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and adjacent submucosal tissues were exposed. During the operation, a mass was found on the sternum. The platysma was found deep in the notch, which was incised before excising the surrounding area. The pathological result is the fourth branchial cleft deformity. After 1 week and 3 months of follow-up, the patients had no discomfort and no recurrence. A review of the relevant literature shows that the fourth branchial cleft deformity is a congenital developmental abnormality that occurs in 1% of all branchial cleft deformity. It often presents as a fistula, cyst, or sinus tract and is anatomically located at the neck root and supravicular region. The fistula is close to the medial lower boundary of the sternocleidomastoid muscle. The diagnosis is often made based on its anatomical location, imaging examinations and, ultimately, pathology. The differential diagnoses include other cervical swellings, such as hemangioma and a thyroglossal duct cyst. Surgical resection is a commonly used treatment method. In recent years, endoscopic positioning and internal fistula burning have had good curative effects for recurrent fourth branchial cleft deformity, with a small chance of recurrence or cancer. **Conclusion** Given its unique position, clinicians should make full use of imaging methods to determine the size, anatomical location and course of the lesion when treating the fourth branchial cleft deformity to ensure the complete and safe surgical resection of the lesion and prevent recurrence.

【Key words】 branchial cleft deformity; fourth branchial cleft; branchial cleft sinus; branchial remnant; pre-sternal region; sternoclavicular joint; cyst

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鳃裂畸形包括鳃裂囊肿、鳃裂窦和鳃裂瘻,起源于鳃裂,根据鳃裂的起源可分为四种类型,其起源多数认为是由胚胎鳃裂残余组织所形成,囊壁厚薄不等,含有淋巴组织,通常多覆有复层鳞状上皮,少数被以柱状上皮。在胚胎发育的第3周时,头部两侧各有5对斜行突起、平行的腮弓,腮弓之间,外侧为凹进的沟形鳃裂所分离,内侧则为凸出的咽囊^[1],而第四腮裂畸形是一种先天性发育性异常,系由正常胚胎结构退化失败引起,根据胚胎发生期间不完全闭塞的程度,常以瘻管、囊肿或窦道存在,极为罕见,在腮裂畸形发病率中占比不到1%,好发于左侧^[2-3]。

本文报告1例口腔科极少见的第四腮裂畸形,并结合相关文献,对其起源、临床表现、诊断、治疗及其预后等进行讨论。

1 临床资料

1.1 一般资料

患者,女,28岁,主诉:左侧前胸壁下肿物反复肿痛2个月,破溃后形成瘻口。患者于2022年1月前来徐州市中心医院就诊,首诊于普外科,普外科请口腔科会诊后收入口腔科进一步治疗。

患者自小时记事起左侧前胸壁周围肿块病史,颈部肿块反复感染史,感冒后加重,2个月前肿

物渐进性增大,遂于外院行脓肿切开引流术,引出黄色清亮液体,后形成瘻口,经久不愈。

既往否认心脏病史,高血压病史以及过敏史等。

专科检查:左侧锁骨下方可见一大小为10 mm×10 mm的瘻口(图1a),瘻口流出黄色清亮的液体。向上胸锁关节浅面颈胸交汇处可触及一大小为20 mm×20 mm×10 mm的肿物,无波动感,活动度欠佳,按压稍疼痛。周围淋巴结未见明显异常。其余组织未见明显异常。

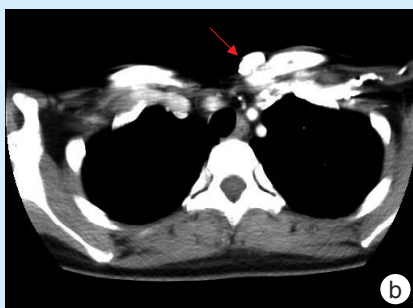
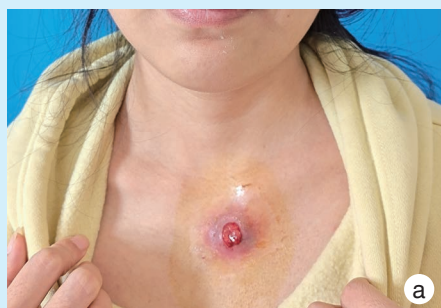
口内检查:全口牙列完整,牙龈、腭部、颊部黏膜未见红肿糜烂。

1.2 相关辅助检查及会诊资料

1.2.1 实验室检查 血常规、电解质及肝肾功能等均无明显异常。

1.2.2 影像学检查 对瘻管处进行造影显影,CT检查示:管状囊状结构沿胸锁乳突肌向左下延伸至锁骨下(图1b),肿物位于皮肤黏膜下。边界不清,周围均匀薄而光滑;颈部超声显示在前胸壁下方有一个囊性病变,可探查至一27 mm×24 mm×9 mm大小低回声,界不清,形态不规整,中央皮下可见少许液性暗区。

1.2.3 耳鼻喉科会诊 耳鼻喉科会诊意见:患者前胸壁肿物,结合颈部CT及超声表现,考虑为第四



a: preoperative frontal photo, the fistula was located under the left clavicle; b: the arrow points to a branchial cleft cyst, which was a tubular cystic structure extending from the lower left to the subclavian along the sternocleidomastoid muscle

Figure 1 Preoperative photo and neck CT of a patient with deformity of the fourth branchial cleft on the left anterior chest wall

图1 左前胸壁第四鳃裂畸形患者术前照片及颈部CT

腮裂畸形,可行手术切除,术后随访观察。

1.3 治疗

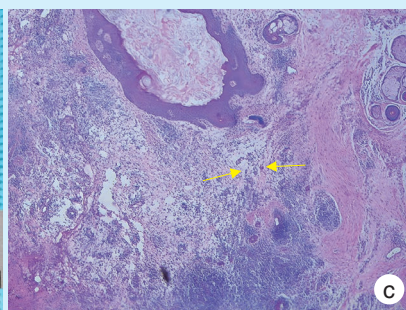
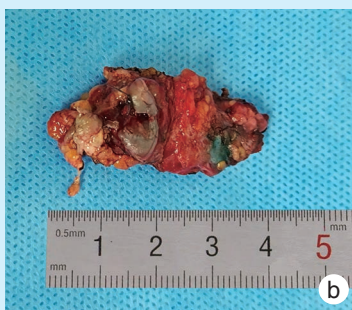
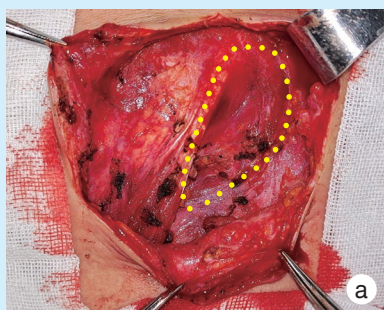
入院后初步诊断为“第四腮裂畸形”,术前予以抗炎消肿等对症治疗。

手术治疗过程:于患者锁骨下瘘口痕迹、触及囊肿处注入少量亚甲蓝,设计锁骨上T型切口,翻瓣显露亚甲蓝染色的所有区域及邻近的黏膜下组织,追溯瘘管沿胸锁乳突肌、胸骨甲状肌到达其深面,上达甲状腺位置,游离上端,切除周围亚甲蓝染色组织,下达胸骨上方、锁骨下缘,切除瘘口及囊肿。术中,肿块很容易在胸骨上切迹颈阔肌深

处找到,并沿其周围切开并切除。再次仔细寻找,未发现亚甲蓝染色的组织(图2a),大量生理盐水冲洗术区,检查无活动性出血,放置负压引流管1根,分层缝合创口,术区加压包扎。

1.4 病理学检查

病理诊断:大体病理检查显示囊肿大小40 mm×30 mm×10 mm(图2b)。切开见囊腔,连续切片显示囊肿内有灰黄乳白色物质,囊腔直径为0.6 cm,鳞状上皮囊肿伴急慢性炎,及多核巨细胞反应。病理结果为:第四腮裂畸形(图2c)。



a: the boundary of the operation area (dotted line) was clear, and it was slightly adhered to the surrounding soft and hard tissues; b: postoperative specimen showed complete excision of fistula and cyst; c: HE staining showed squamous cyst with acute and chronic inflammation and multinucleated giant cell reaction (yellow arrows) (×100)

Figure 2 Intraoperative, surgical specimens and postoperative pathology of a patient with deformity of the fourth branchial cleft on the left anterior chest wall

图2 左前胸壁第四鳃裂畸形术中、术后标本及术后病理

1.5 术后观察随访

常规换药,术后3 d拔除引流管,术后7 d拆除术区缝线,创口愈合可,出院后随访1周、3个月无复发,预后可(图3)。

2 讨论

本病例描述了一个位于不寻常位置的第四腮裂畸形,通过文献检索,此类腮裂畸形在口腔领域不仅国外很少报道,国内的文献也大都以个案的



a: the wound healed and there was no infection 1 week after the surgery; b: the wound basically healed without recurrence 3 months after the surgery

Figure 3 Follow-up photos of a patient with deformity of the fourth branchial cleft on the left anterior chest wall 1 week and 3 months after surgery

图3 左前胸壁第四鳃裂畸形患者术后1周及3个月随访照片

形式出现^[4],2016年黄舒玲等^[5]报道了开口于胸锁关节浅面颈胸交汇处皮肤的鳃源性窦道,近几年未出现过类似报道。值得一提的是在口腔科目前较少发现有对于第四鳃裂畸形的报道,罕见的原因可能有:①第四鳃裂畸形本身的罕见性;②鳃裂畸形属于口腔医学与耳鼻喉科学的交叉学科,但是因第四鳃裂畸形的位置特殊,患者大多去耳鼻喉科就诊。因此该病例对口腔科医生诊断并治疗鳃裂畸形,提供了重要的参考价值。

第一鳃裂畸形约占所有鳃裂畸形的5%~25%,第二鳃裂畸形占鳃裂畸形的40%~95%^[6];第三和第四鳃裂畸形是一种罕见的先天性颈部异常,尤其是第四鳃裂畸形极为罕见,约占所有鳃裂畸形的1%^[3]。第四鳃裂畸形的特征是它们多位于左侧,皮肤开口靠近胸锁乳突肌的内侧下边界,其明确的原因仍不清楚,可能与第四腮弓以及血管结构的不对称发育有关。相比较于左侧,右侧更为少见,Meng等^[7]报道了一例发生在右侧的位于胸骨柄和胸骨前方的罕见第四鳃裂畸形。

第四鳃裂畸形的解剖学位置位于颈根部、锁骨上区,可与甲状腺相毗邻的囊肿或窦道,可起源于一侧梨状窝,经环杓关节迂曲走行于喉上神经和喉返神经之间,反复感染时伴有甲状腺炎或甲状腺脓肿^[8-9]。第四鳃裂瘘管外口的解剖位置与第二、三鳃裂瘘管相同或在胸前部经颈阔肌深面、沿颈动脉鞘下行至胸部,皮肤开口靠近胸锁乳突肌的内侧下边界。

在大多数情况下,鳃裂畸形无自觉症状。从新生儿期到成年期,第四鳃裂畸形可以多种不同方式出现。新生儿由于鳃裂畸形导致气道狭窄,主要表现为呼吸窘迫^[10]。在较大的儿童和成人中,最常见的表现为复发性脓肿或伴有颈部感染的皮肤溢液瘘^[11],还有可能出现复发性上呼吸道感染、复发性急性化脓性甲状腺炎、颈部或甲状腺

疼痛、颈部肿块,或者更罕见的伴有脓肿、蜂窝组织炎、吞咽困难以及吞咽痛或声音改变^[12-13]。

第四鳃裂畸形常由于误诊和随后的复发,病损经历了反复的切开引流等手术治疗导致瘢痕形成,使诊断更加困难;发生在颈部的肿物缺乏特异性症状,临床易误诊或漏诊,大多数表现为无痛性颈部肿块,并反复感染,尤其是上呼吸道感染时加重。由于病变局部反复感染甚至破溃,使囊肿十分难以辨识,因此不应排除颈部肿块如:淋巴结病、血管瘤、颈动脉体瘤、异位甲状腺/唾液组织、甲状舌管囊肿以及囊性鳞状细胞癌等^[14]进行鉴别诊断。本病例中患者瘘管存在2个月,因此鳃裂畸形里的囊液残留较少,且持续流出黄色清亮液体,因此囊肿不明显,只可见一明显瘘管,囊肿经反复感染后经胸前壁锁骨下破溃形成瘘口,经久不愈。第四鳃裂畸形本身就较罕见,瘘口位于胸前壁锁骨下方更是少见,进行鉴别诊断时,主要是通过该病的术后病理作为与胸腺囊肿^[15]的主要鉴别。

鳃裂畸形通常是良性的,手术切除历来被认为是鳃裂畸形的最佳治疗方法。查阅国内外文献,较罕见的第四鳃裂畸形的治疗近年一直在不断研究中。选择性颈淋巴清扫术能显著控制早期口腔鳞状细胞癌颈部淋巴结转移率^[16],Ning等^[17]报道发现选择性颈淋巴清扫术治疗多次复发或反复颈部感染的第四鳃裂畸形安全有效;当存在明显的甲状腺受累时,第四鳃裂畸形的联合治疗包括内镜烧灼、手术完全切除和半甲状腺切除术,也是安全有效的^[18];针对于婴幼儿及儿童患者,通常选择微创治疗,在内窥镜下烧灼来消除梨状窦道的内部开口,Saadoun等^[19]也报道可在内窥镜下对第四鳃裂畸形内瘘口开口梨状窝进行电凝手术,以防止复发;当有复发性感染和靠近喉返神经束时,也建议将通过内镜烧灼内瘘口结合手术彻底切除作为第四鳃裂畸形的首选治疗^[20]。为了防止

复发,对于异常的内口,缝合结扎烧灼比单纯烧灼的复发率低,这种方法对难治性病例有益^[21]。

本病例中,术前通过超声并结合影像学方法明确病变的大小、部位及走行,通过手术治疗即可完整、安全切除病灶,术后及时回访,对于预防病灶复发具有重要意义。通过治疗,患者很快康复,没有出现并发症或复发,并且本病中瘻管通道没有穿过甲状腺,患者也没有出现化脓性甲状腺炎等的症状,因此没有进行甲状腺部分切除术。因为有感染、进一步扩大或恶化的风险,在手术前进行超声、CT或MRI检查是必要的,这些检查可以引导手术进行。如超声可以确定囊肿特征,CT有助于明确肿物的位置和病变界限;如果存在窦道,可以通过注射造影剂来获得造影;手术时建议用亚甲蓝注射使瘻道显色,以明确走行并确定囊肿大小。在本病例中不足的一点是通过增强CT来显影,没有X线清晰,因此建议后来研究者造影时使用X线检查^[22]。

良性的腮裂畸形通常经过手术完整切除后一般具有良好的预后,其最常见术后并发症为手术部位的感染^[23];腮裂畸形少数可发展为鳃裂癌^[24-25],但发病率较低,当进行鳃裂癌的诊断时,建议在手术前通过影像学资料来区分腮裂畸形良、恶性^[26];有研究者报道了一例发生于腮腺区的第一鳃裂癌,经过手术治疗后预后较好,基于病例数较少,指出当伴有颈部淋巴结转移时,是否比其他头颈部鳞状细胞癌有更好的预后则有待于更多的病例积累和临床随访^[27];虽然鳃裂癌治疗目前尚没有统一标准,但是早发现、早诊断和早治疗始终是提高鳃裂癌患者治愈率及生存率的关键。

综上所述,针对患者的不同情况形成第四腮裂畸形的个性化治疗模式正在不断探索中,随着第四腮裂畸形的病例报道增多,未来有望形成更加规范的治疗流程和临床路径。

【Author contributions】 Zhao S, Tang JL collected case materials and wrote the article. Shen MY, Kang N revised the article. Li XD, Meng J were the performer and analyzed the case. All authors read and approved the final manuscript as submitted.

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官网

• 短讯 •

《口腔疾病防治》被日本科学技术振兴机构数据库(JST)收录

近期编辑部获悉,由南方医科大学口腔医院主办的《口腔疾病防治》被日本科学技术振兴机构数据库(Japan Science & Technology Corporation, JST)收录。

日本科学技术振兴机构数据库(JST)(原日本科学技术社数据库)是在日本《科学技术文献速报》(CBST,被誉为世界六大著名检索系统)的基础上发展起来的网络版数据库,隶属于日本政府文部科学省的科学技术管理组织。JST检索源期刊的遴选采取独立评选方式,不接受推荐期刊。

这是继《口腔疾病防治》被美国《乌利希期刊指南》(Ulrichsweb)、荷兰Scopus数据库、瑞典《开放获取期刊指南》(Directory of Open Access Journals, DOAJ)、波兰《哥白尼索引》(Index Copernicus, IC)、瑞士《健康网络首创研究获取》(Health InterNetwork Access to Research Initiative, HINARI)、WHO西太平洋地区医学索引(The Western Pacific Region Index Medicus, WPRIM)等国际重要数据库收录后入选的第7个国际重要数据库。这标志着本刊学术质量和国际影响力的再次提升,并得到了国际相关学科组织的认可。在此,本刊编辑部向长期关心、支持《口腔疾病防治》发展的各级领导、全体编委、审稿专家、广大作者、读者致以衷心的感谢!

《口腔疾病防治》编辑部