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· 防治实践 ·

同侧腮腺原发2种不同肿瘤1例及文献回顾

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【摘要】 目的 探讨同侧腮腺内同时原发不同病理类型肿瘤的临床表现、组织病理学特点、诊断治疗及预后。方法 报道1例同侧腮腺内腺淋巴瘤和基底细胞腺瘤同时发生的病例,并结合文献对腮腺多原发肿瘤的诊疗进行回顾分析。结果 患者发现右腮腺区包块1个月,术前扪及右腮腺浅叶肿物,结合核磁共振检查诊断为右侧腮腺浅叶及深叶肿瘤。全麻下行右侧腮腺深叶及浅叶肿物和部分腺体切除术+面神经解剖术,术后病理报告右侧腮腺浅叶为腺淋巴瘤,深叶为基底细胞腺瘤伴囊变。手术效果良好,无并发症,术后随访1年未见复发。复习文献结果表明,腮腺多原发性肿瘤是指双侧或者单侧同时存在2种或以上的肿瘤。该疾病以手术治疗为主。结论 同侧腮腺内同时原发不同病理类型的肿瘤病例罕见,需完善影像学检查防止漏诊,手术为首选治疗方案,良性肿瘤预后良好。

【关键词】 腮腺; 多原发性肿瘤; 腺淋巴瘤; 基底细胞腺瘤; 细针穿刺活检; 磁共振成像; 术中冰冻病理; 手术入路; 包膜外切除

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Unilateral parotid gland Simultaneously primary two different pathological tumors: a case report and literature review XIE Hongliang, TANG Jianming, ZHANG Guoquan. Department of Oral and Maxillofacial Surgery, Stomatological Medical Center & Shenzhen People's Hospital & the First Affiliated Hospital of South University of Science and Technology & the Second Clinical Medical College of Ji'nan University, Shenzhen 518020, China

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【Abstract】 Objective To explore the clinical manifestations, histopathological characteristics, diagnosis, treatment, and prognosis of simultaneous unilateral primary tumors of different pathological types in the parotid gland. **Methods** A case of simultaneous unilateral primary parotid gland tumors, i.e., adenolymphoma and basal cell adenoma, was reviewed and analyzed in combination with the literature. **Results** The patient discovered a lump in the right parotid gland area one month prior to presentation, and a tumor was palpated in the shallow lobe of the right parotid gland before surgery. According to MR images, the initial diagnoses were tumors of the shallow and deep lobes of the right parotid gland. The tumors of the deep and shallow lobes were excised with part of the gland, and the facial nerves were dissected under general anesthesia. Postoperative pathology revealed an adenolymphoma in the shallow lobe of the right parotid gland and a basal cell adenoma with cystic transformation in the deep lobe. The surgical effect was good, with no complications, and there was no recurrence after 1 year of follow-up. A review of the relevant literature showed that multiple primary tumors of the parotid gland can manifest as the simultaneous presence of two or more types of tumors on both sides or on one side, and the disease is mainly treated with surgery. **Conclusion** Multiple unilateral primary parotid gland tumors are rare. Imaging examinations need to be combined with clinical evaluations to prevent missed diagnoses. Surgery is the first treatment option, and patients with benign tumors have a good prognosis.

【Key words】 parotid gland; multiple primary tumors; adenolymphoma; basal cell adenoma; fine needle aspira-

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tion biopsy; magnetic resonance imaging; intraoperative frozen pathology; surgical approach; extracapsular resection

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多原发性肿瘤(multiple primary tumor, MPT)是指在同一宿主单个器官或者多个器官同时或者先后发生的2个或者2个以上的原发性肿瘤^[1-2]。根据肿瘤发生时间,可分为同时原发和异时原发,其中异时原发里先发现确诊的为先证肿瘤(index tumor)即第一原发肿瘤,后发现确诊的为第二原发肿瘤。腮腺多原发性肿瘤是指双侧或者单侧同时存在2种或以上的肿瘤。根据肿瘤发生的位置,可分为单侧多发、双侧单发及双侧多发等^[3]。本研究报道1例同侧腮腺内腺淋巴瘤和基底细胞腺瘤同时发生的病例,探讨同侧腮腺内同时原发不同病理类型肿瘤的临床表现、组织病理学特点、诊断治疗及预后。

1 同侧腮腺原发2种不同肿瘤典型病例

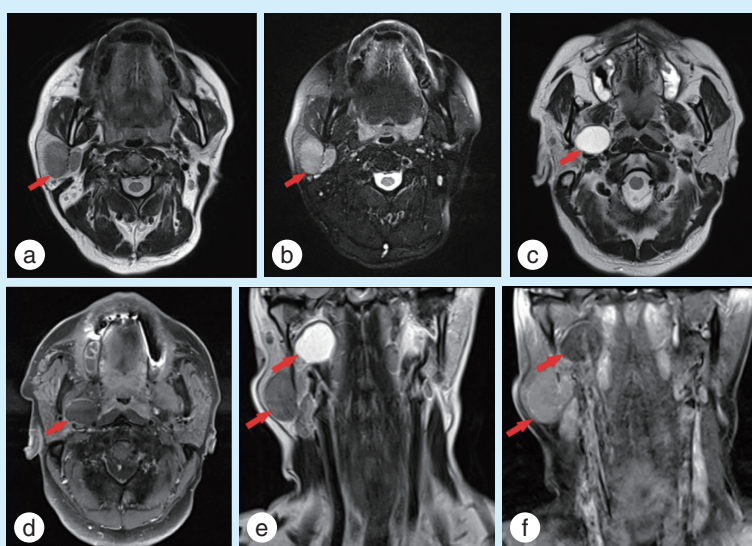
1.1 初诊资料

患者,男,54岁,2022年5月因发现右腮腺区无痛性包块1个月入院,现病史:1个月前无意中

发现右腮腺区耳垂后下方处有一小肿物,约鹌鹑蛋大小,无疼痛不适,在当地医院就诊行B超示:右腮腺内实质性占位病变。后来我院就诊,以“右腮腺腺淋巴瘤?”收入院。既往史:否认高血压、心脏病、糖尿病等系统性疾病,否认传染病病史,否认输血史,否认食物药物过敏史,常规预防接种史。个人史:吸烟30年,平均每日吸烟20支。专科检查:右腮腺耳垂后下方可扪及一直径约4 cm肿物,质地中等偏软,活动度可,无触压痛,边界清,无面神经受侵犯表现。辅助检查:磁共振成像示右侧腮腺见类圆形长T1长T2信号结节,大小约27 mm × 26 mm,边界清楚,呈分叶状,累及右侧腮腺浅叶。增强后病灶轻度持续强化。右侧腮腺深叶内侧与翼外肌之间见一囊性长T1长T2信号,大小约25 mm × 17 mm,增强扫描内部未见强化,边缘环形强化(图1)。

1.2 治疗方案

患者入院后完善术前相关检查,排除明确手



a: well-defined lesion about 27 mm × 26 mm located in the shallow lobe of the right parotid gland (arrow showing tumor); b: continuous enhancement of the parotid gland shallow lobe lesions after enhancement; c: a cystic lesion about 25 mm × 17 mm can be seen in the deep lobe of the right parotid gland (arrow showing tumor); d: after enhancement, there is no internal enhancement in the deep lobe lesion of the parotid gland, with circular enhancement at the edge; e: coronal position image simultaneously showed both deep and shallow lobe tumors of the right parotid gland; f: coronal position enhancement image showed different enhancement characteristics of deep and shallow lobe tumors in the right parotid gland

Figure 1 Enhanced magnetic resonance imaging results of the maxillofacial region in a 54-year-old male patient with unilateral parotid gland simultaneously primary two different pathological tumors

图1 54岁男性患者右侧腮腺原发2种不同肿瘤颌面部增强核磁共振成像检查

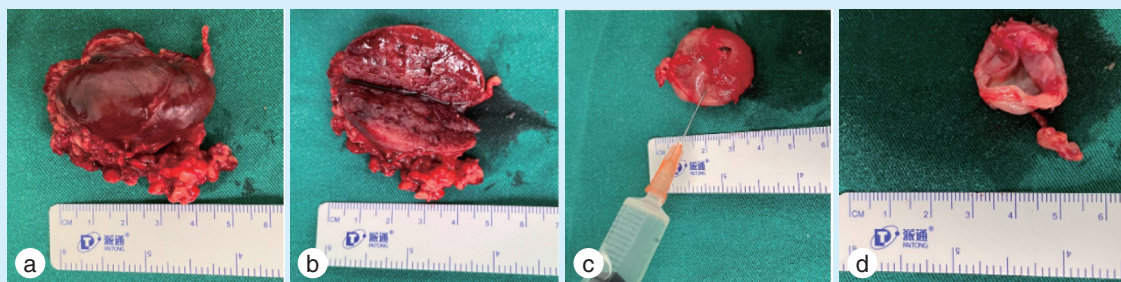
术禁忌证,于全麻下行“右侧腮腺深叶及浅叶肿物和部分腺体切除术+面神经解剖术”,手术入路为经典耳屏前绕耳垂至颌后下颌角下“S”形切口,翻开腮腺咬肌筋膜,解剖并保护面神经分支及主干后切除部分腮腺浅叶及肿物,然后在面神经下方暴露腮腺深叶肿物并完整切除。

2 治疗结果

手术过程顺利,术中面神经保存完好,完整切除2个肿瘤,见腮腺浅叶肿瘤大小4.5 cm × 2 cm × 2 cm,肉眼可见包膜完整,剖面灰红实性质软;深叶肿瘤包膜完整,囊性,大小2.8 cm × 2 cm × 1 cm,壁厚0.2 cm,内含清亮稀薄液体(图2)。

术中冰冻病理提示:右腮腺浅叶肿物:腺淋巴瘤(Warthin瘤),周围触及淋巴结1枚,呈反应性增生。右腮腺深叶肿物:基底细胞腺瘤。术后患者无面瘫、无涎瘘等并发症。术后石蜡病理报告:右侧腮腺浅叶肿瘤考虑为腺淋巴瘤(Warthin瘤),肿瘤由腺管及淋巴间质构成,周围触及淋巴结1枚,呈反应性增生,深叶肿瘤考虑为基底细胞腺瘤伴囊性变,囊壁由基底膜和基底样上皮细胞组成,囊壁部分形成小管状结构,包含两层细胞,内层为柱状细胞和鳞状细胞,外层为肌上皮细胞,细胞体积较小,细胞质较少,染色较深,小管内包含嗜伊红物质(图3)。

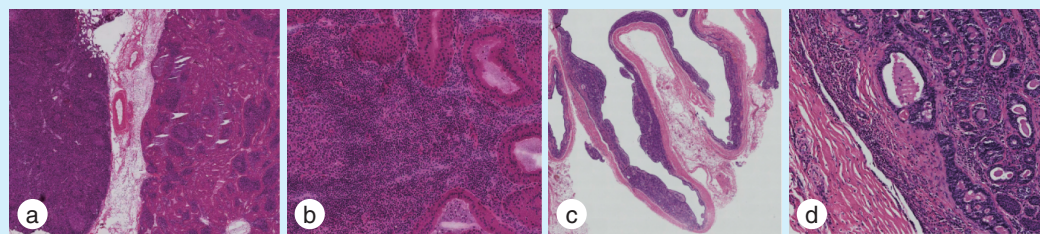
术后随访1年,复查腮腺B超,未见肿瘤复发。



a: the capsule of the right parotid gland shallow lobe tumor is intact, with a maximum diameter of approximately 4.5 cm; b: the tumor in the shallow lobe of the right parotid gland is mainly solid in profile, with small cystic changes visible and containing dark red fluid; c: the deep lobe tumor of the right parotid gland is cystic, with a diameter of approximately 2.5 cm and contains clear, thin, and transparent liquid; d: uneven thickness of the cyst wall in the deep lobe of the of two tumors removed from patient with unilateral parotid gland simultaneously primary two different pathological tumors right parotid gland

Figure 2 Appearances of two tumors removed from a 54-year-old male patient with unilateral parotid gland simultaneously primary two different pathological tumors

图2 54岁男性患者右侧腮腺原发2种不同肿瘤切除肿瘤的外观



a: the tumor of the the right parotid gland shallow lobe presents as a typical adenolymphoma, with reactive hyperplasia in the adjacent lymph node (HE×10); b: the tumor in the shallow lobe of the right parotid gland consists of glandular ducts and lymphoid stroma(HE×100); c: the deep lobe tumor of the right parotid gland is cystic, and its wall is composed of basement membrane and basal like epithelial cells(HE×10); d: the wall of the deep lobe tumor of the right parotid gland forms a tubular structure, which contains two layers of cells, the inner layer is columnar cells and squamous cells, the outer layer is myoepithelial cell, and the tubule contains eosinophil (HE×100)

Figure 2 Pathological results of two tumors removed from a 54-year-old male patient with unilateral parotid gland simultaneously primary two different pathological tumors

图3 54岁男性患者右侧腮腺原发2种不同肿瘤切除肿瘤的病理结果

3 讨论

腮腺肿瘤多为单发,多原发性腮腺肿瘤发病率极低,约占腮腺全部肿瘤的3.4%,多见于老年男性。其中最常见的多原发肿瘤病理类型是腺淋巴瘤,约占80%,这可能是因为腺淋巴瘤本身具有多发中心的特点;其次为多形性腺瘤^[4]。而同时发生于同侧腮腺不同病理类型的病例较为罕见,多为个案报道^[5-6]。

不同病理类型的肿瘤来源不同,无相关性,因此,多数学者认为,除了多发腺淋巴瘤,其余类型的腮腺多原发肿瘤是一种巧合现象^[7],发生率大约为13%。腺淋巴瘤又称为Warthin瘤,是腮腺第二高发的良性肿瘤,最近有多中心研究发现其发病率有上升趋势,超过多形性腺瘤^[8]。腺淋巴瘤是临床最常见的多发性唾液腺肿瘤,多见于老年男性,且患者多有吸烟史,而腺淋巴瘤合并其他肿瘤的比例也明显高于其他病理类型的多原发性肿瘤^[9]。

关于腺淋巴瘤多发性的发病机制,最被学者认可的假说是在胚胎发育时期由存在于腮腺淋巴结内的腮腺组织发生而来。在胚胎发育的早期阶段,腮腺腺体内上皮和淋巴组分之间尚无明显界限,此时腺体上皮细胞与腺导管腺泡系统前体有一部分嵌入在淋巴组分内,而这些淋巴组分在发育过程中形成腺体内淋巴结。此后,在外在刺激因素(例如吸烟)下,这些淋巴结内的上皮细胞成分就有可能形成腺淋巴瘤^[10]。本例患者腺瘤是比较少见的腮腺肿瘤,在组织学上被归为单形性肿瘤,主要由基底细胞构成,并可见基底膜。目前其发病机制仍不明确,一些外在刺激因素例如吸烟、辐射、病毒感染等均可能导致唾液腺肿瘤的发生^[11]。本例患者腮腺深叶的肿瘤与浅叶肿瘤相隔较远,各自发生,影像学表现及镜下表现也证明了这一点。

腮腺多原发肿瘤具有不同时间不同部位发生的特点,因此临床常易漏诊。临床体格检查对位于腮腺浅叶的肿物基本可以判断,但对于腮腺深叶的肿物无法准确判断,容易遗漏。B型超声多普勒检查因其方便快捷无创^[12],临床应用较为广泛,对肿瘤的数量、大小、边界等情况可做观测,能大致辨别淋巴结与肿瘤,并有助于术前定位腮腺浅叶和深叶肿瘤,可帮助确定合适的手术方案^[13]。不过,超声检查对肿瘤三维显示不足,对深叶肿瘤的判断有限。计算机断层成像(CT)或核磁共振成像(MR)则可清晰显示肿瘤与周围组织的关

系^[14-16],尤其是MR,对软组织的分辨率更好,适合腮腺肿瘤,尤其是对腮腺深叶肿瘤的检查诊断具有极为重要的价值^[17-18]。本病例临床体格检查仅触及腮腺浅叶肿瘤,考虑腺淋巴瘤,后完善MR检查发现腮腺深叶肿瘤。术中发现腮腺深叶肿瘤位于深叶深面,需翻开深叶腺体才能暴露。由此可见,若不进行CT或MR检查,腮腺深叶肿瘤很容易发生漏诊、手术不彻底的情况。因此,对于腮腺肿瘤,推荐完善CT最好是MR检查,排除深叶或对侧肿瘤性病变。同时,通过CT或MR可判断肿瘤的位置、与周围血管神经的关系,初步判断肿瘤性质,为临床手术提供指导。

研究报道基底细胞腺瘤有近70%可发生囊变^[19],本例患者腮腺深叶肿瘤为中央大片状囊变。基底细胞腺瘤缺乏黏液样和软骨样物质,这可与多形性腺瘤相鉴别。

手术治疗目前还是作为腮腺多原发性肿瘤治疗的首选。大部分腮腺肿瘤为良性,多采用腮腺肿瘤切除、保留面神经的腮腺浅叶或全叶加肿瘤切除手术。根据肿瘤所在的部位以及良恶性采取相应的手术方案:位于浅叶的良性肿瘤,切除浅叶腺体和肿瘤组织;深叶的肿瘤,则行全腺叶切除手术;对于腮腺恶性肿瘤,不管位于浅叶还是深叶均采取全叶切除,必要时需辅助术后放疗,减少复发率,提高生存率。关于术前穿刺活检明确肿瘤性质目前仍存在争议,有学者认为术前细针穿刺活检可能会导致恶性肿瘤细胞的种植和转移,而且其假阳性或者假阴性率都较高;也有学者则认为虽然细针穿刺活检有一定的局限性,但其在区分肿瘤良恶性及鉴别诊断中具有重要价值^[20-21]。本病例不做术前穿刺,直接术中做快速冰冻病理明确肿瘤性质。该病例术中快速冰冻提示为腺淋巴瘤和基底细胞腺瘤,均为良性。腺淋巴瘤易多发,肿瘤周围淋巴结需一并切除^[22];本例患者浅叶肿瘤周围淋巴结予以一并切除,病理提示淋巴结反应性增生。基底细胞腺瘤切除术后复发率低,而多形性腺瘤可能包膜不完整,手术不彻底者易复发^[23]。因此对于腮腺多原发性良性肿瘤,建议行腮腺区域性切除、浅叶切除或全叶切除,避免因肿瘤残存或肿瘤细胞种植引起复发^[24]。

良性肿瘤术后面瘫发生率约5.4%,恶性肿瘤术后面瘫发生率约19.4%^[25]。本例患者术后未出现面瘫症状。近年来有越来越多的学者主张功能外科,即对于直径小于4 cm的腮腺单发良性肿瘤

可选择包膜外切除术,该技术降低了Frey综合征(味觉出汗)和手术部位凹陷的风险,也可以降低面神经损伤的风险^[26]。尤其对于单发的腺淋巴瘤,有研究显示行肿瘤摘除术即可获得很好的治疗效果,复发率低,而且术后并发症少,微创、瘢痕小^[27]。本病例术中冰冻提示浅叶为腺淋巴瘤,深叶为基底细胞腺瘤,采取的手术方式为部分浅叶及肿瘤切除,深叶肿瘤做包膜外切除,手术效果满意,术后未出现面瘫、涎瘘等并发症,局部凹陷也不明显。

关于腮腺手术切口的选择,传统经典的“S形切口”又称为Blair切口,该切口术野暴露广泛清楚,适用于腮腺各区域的肿瘤,本病例因肿瘤涉及腮腺浅叶和深叶,采用的即为此切口,但该切口局部瘢痕较明显。近年来在美容外科理念的影响下,有学者主张根据腮腺肿瘤的位置、性质、大小、深度,并综合考虑患者的年龄、性别、美观要求等因素设计不同的改良切口,以达到在治疗肿瘤的同时满足患者的美观需求,并进行研究和临床手术改进,取得良好的效果^[28]。

综上,对于腮腺肿瘤,术前应尽量完善CT或者MR检查,排除一侧或双侧多发情况,防止腮腺深叶肿瘤遗漏。术中对肿瘤周围的淋巴结一并切除送冰冻切片检查,若淋巴结提示为肿瘤病变,则需适当扩大手术,切除周围腺体组织及淋巴结,降低术后复发率。对于腮腺恶性肿瘤,则需要手术切除全部腺体,部分恶性程度高的需要同期行颈部淋巴结清扫术联合术后放化疗,提高生存率。

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