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同侧上下颌骨单纯性骨囊肿1例报告及文献复习

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[摘要] **目的:** 分析同侧上下颌骨单纯性骨囊肿(SBC)患者的临床表现、影像学特征、手术探查情况和病理学表现, 以提高临床医生对该类疾病诊断和治疗的认知。**方法:** 收集1例右侧上下颌骨SBC患者的临床资料、影像学特征、手术探查情况、病理学表现、临床诊断和治疗经过, 并结合相关文献进行复习。**结果:** 患者, 男性, 11岁; 曲面断层检查, 发现右侧上下颌骨内边界清楚的低密度影; 无创伤史, 就诊前无自觉症状和面部不对称; 专科检查未见任何阳性体征; 结合手术探查情况及病理学诊断确诊为右侧上下颌骨SBC; 采用传统刮治术治疗, 术后患者恢复良好, 无明显不适。术后6个月随访, 患者口内创口愈合良好, 无红肿; 锥形束计算机断层扫描(CBCT)影像检查结果显示骨腔变小, 骨腔内可见新生骨小梁, 成骨情况良好, 目前患者处于定期随访中。**结论:** 同侧上下颌骨SBC无特征性的临床表现和影像学表现, 极易与颌骨常见的疾病混淆。需结合影像学检查、手术探查情况和病理学检查最终确诊。

[关键词] 创伤性骨囊肿; 单纯性骨囊肿; 孤立性骨囊肿; 刮治术; 病例报告

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Simple bone cyst in ipsilateral maxilla and mandible: A case report and literature review

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ABSTRACT Objective: To analyze the clinical presentations, radiographic features, operative findings, and pathological characteristics of one patients with unilateral maxillary and mandibular simple bone cysts (SBC), and to enhance the clinicians' recognition and treatment of this condition. **Methods:** The clinical data, radiographic features, operative findings, pathological characteristics, clinical diagnosis, and treatment of a case with right-sided maxillary and mandibular SBC were collected, and the relevant literatures were reviewed. **Results:** The patient, an 11-year-old male, presented with a clearly demarcated low-density image within the right maxillary and mandibular bones on panoramic tomography. There was no history of trauma, no subjective symptoms or facial asymmetry before treatment, and no positive signs of the specialist examination. The patient was diagnosed with right-sided maxillary and mandibular SBC based on the operative exploration and pathological diagnosis. The patient received the conventional curettage treatment and recovered well postoperatively without significant discomfort. A

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6-month follow-up results showed good intraoral wound healing without swelling. The cone beam computed tomography (CBCT) results showed a smaller bone cavity with visible new bone trabeculae, indicating good osteogenesis. The patient was currently the under regular follow-up. **Conclusion:** Unilateral maxillary and mandibular SBC do not present with characteristic clinical signs and radiographic features, which can be easily confused with the common maxillofacial diseases. A definitive diagnosis should be made by combining radiographic examination with surgical and pathological findings.

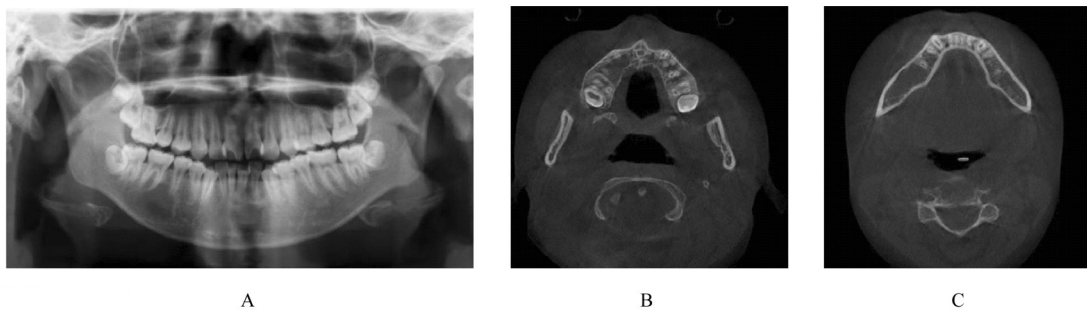
KEYWORDS Traumatic bone cyst; Simple bone cyst; Solitary bone cyst; Curettage; Case report

颌骨单纯性骨囊肿 (simple bone cyst, SBC) 是发生于颌骨内的良性病变。单纯性骨囊肿常见于青少年四肢长骨中, 发生于颌骨中的 SBC 较为少见, 上下颌骨内多发的 SBC 病例则更为罕见^[1-4]。下颌 SBC 多发生于后牙区, 也有少部分发生于颞下颌关节和下颌升支, 上颌 SBC 多发生于上颌前部^[5-10]。SBC 发病无性别差异, 且在 10~20 岁更易发生, SBC 患者常无临床症状, 波及牙的牙髓活力测试正常, 部分患者可并发面部肿胀、疼痛及牙痛^[2, 11-12]。目前, 关于同侧上下颌发生 SBC 的患者尚不多见。本文作者探讨 1 例 SBC 患者的临床资料、影像学特征、手术和治疗方案及病理形态表现, 结合相关文献复习, 为临床明确诊断该类疾病并选择合适的治疗方案提供参考。

1 临床资料

1.1 病例资料 患者, 男性, 11 岁; 2021 年 8 月

因牙列不齐行正畸治疗于本院正畸科就诊, 拍摄曲面断层片检查时发现右侧上下颌骨内有 2 处类圆形低密度骨缺损, 患者无不适症状。患者否认牙痛及创伤史, 无家族史; 专科检查未见病变区的阳性体征; 口内 14-17 和 34-37 牙无龋坏、充填物、叩痛、松动及移位。右侧上颌和下颌未触及骨质膨隆, 按压无痛; 锥形束计算机断层扫描 (cone beam computed tomography, CBCT) 检查结果显示: 右上颌骨前磨牙区可见类圆形高密度影, 边界清楚, 无骨白线, 大小约为 1.8 cm×1.2 cm, 14 和 15 牙根位于病变内, 病变累及 13 腭侧骨质。右下颌磨牙区可见类圆形骨质密度透光区, 边界清楚, 大小约为 2.8 cm×2.0 cm, 46 牙根尖位于病变内, 45 和 47 牙根尖距病变仅有薄层骨质相隔或局部相通, 病变累及右下颌神经管 (图 1)。



A: Panoramic radiograph; B: CBCT of maxilla; C: CBCT of mandible.

图 1 同侧上下颌骨 SBC 术前曲面断层片及 CBCT 影像学表现

Fig. 1 Preoperative panoramic radiograph and CBCT images of ipsilateral maxilla and mandible SBC

1.2 治疗方法 患者及其家属均知情同意并签署知情同意书, 全麻下行上下颌骨囊肿摘除术; 术中去除上颌骨病损区表面骨皮质后, 暴露骨创腔, 无内容物及囊壁, 而且囊腔内易出新鲜血 (图 2); 压迫止血后, 搔刮骨创腔, 仅有极少量的软组织, 留送病理活检; 搔刮创腔周围骨壁, 使新鲜血充满骨创腔, 复位黏膜瓣, 严密关闭创口; 采用相

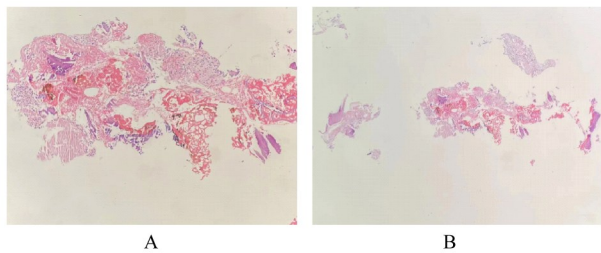
同操作处置下颌骨处病损, 将下颌骨创腔内搔刮的极少量软组织行病理活检; 术后病理检查结果显示: 光镜下可见出血和渗出的纤维蛋白及少量骨组织 (图 3)。病理诊断结果为右上颌骨 SBC。

1.3 术后随访 患者术后康复良好, 无不适或疼痛的症状。进行手术后的 6 个月内进行定期随访, 口内创口愈合良好, 无红肿。CBCT 影像检查结果



图2 术中SBC患者口内图像

Fig. 2 Intraoperative intraoral diagram of SBC patient

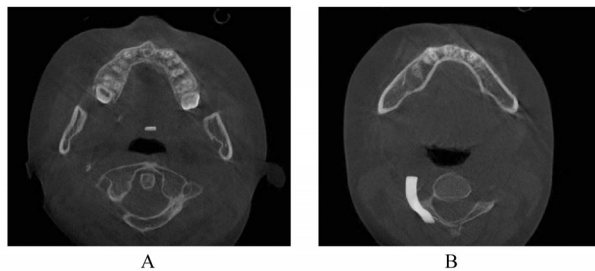


A: Low magnification clens (×40); B: High cmagnification clens (×200).

图3 HE染色观察SBC患者右侧上下颌骨SBC组织形态表现

Fig. 3 Morphology of SBC tissue in right maxilla and mandible of SBC patient observed by HE staining

显示骨腔变小, 骨腔内可见新生骨小梁, 成骨情况良好, 目前患者处于定期随访中 (图4)。



A: CBCT of maxilla; B: CBCT of mandible.

图4 SBC患者同侧上下颌骨术后随访CBCT影像

Fig. 4 Postoperative follow-up CBCT images of ipsilateral maxilla and mandible of SBC patient

2 讨论

目前对于SBC的定义仍在不断完善, 1946年提出的SBC定义标准为一种单纯的骨囊肿, 无内衬上皮, 含有液体, 无软组织, 受骨壁限制, 无感

染迹象^[13-14]。2017年世界卫生组织 (World Health Organization, WHO) 将SBC定义为缺乏上皮衬里或充满浆液及血液的骨内假性囊肿^[15]。

SBC无上皮衬覆的囊壁, 其骨腔内中空或充满浆液及血液, 有一薄层结缔组织膜覆盖于骨表面, 仅在显微镜下可见^[16-17]。目前, SBC的病因和发病机制尚未完全阐明。在其发病机制的多种理论中有3种理论占据主导地位: ①骨生长异常; ②肿瘤退化过程; ③引发出血性创伤的特定因素^[8]。多数学者^[18]认为SBC发病与创伤有关。创伤理论认为: 创伤引起血栓或动脉痉挛, 引发局部缺血和无菌性坏死。研究^[11]显示: 取囊腔面的骨组织进行组织病理学检查, 可观察到充血的毛细血管, 并可见破骨细胞和发育不良的新骨形成。但由于大部分临床报道的病例并无创伤史, 因此仅提示创伤是SBC的致病因素之一^[19]。除外部创伤外, 咬合创伤也可能是SBC的致病因素。此外, 有学者^[20-21]认为: SBC可能与骨内血管畸形有关, 也可能与正畸治疗有关。

大部分颌骨SBC患者无特异性临床表现, 也有部分SBC患者并发纤维结构不良和牙骨质-骨结构不良等其他疾病^[22]。SBC患者的X线和CT影像检查结果显示: 颌骨内有边界清楚的单房性透光影, 少部分为多房性, 无边缘骨白线, 多表现为圆形、卵圆形或不规则形, 在牙根间者表现为扇形^[11]; 病变范围大小不一, 病变范围较大者骨皮质受压变薄, 但无病理性骨折; 受累牙根一般移位且无吸收, 硬骨板仍然存在^[9], SBC的影像学表现与单囊性成釉细胞瘤、慢性根尖周炎、牙源性角化囊肿和中央型巨细胞肉芽肿等疾病的影像学表现极为相似, 难以鉴别^[17]。由于SBC缺乏上皮衬里, 骨腔内压力低, 其液体成分被骨壁上的血管和淋巴管吸收, 在低压力环境下通过破骨细胞的作用使松质骨被吸收, 周围缺乏矿物质沉积, 无法产生硬化边缘, 故影像学检查显示未产生骨白线^[23]。对于类似颌骨囊肿的影像学表现无边缘骨白线的患者, 临床医生应注意其是否为SBC。对于疾病不能确诊者, 可考虑行磁共振成像 (magnetic resonance imaging, MRI) 检查, 判断骨腔内容物性质。由于SBC患者病理资料较少, 故对SBC的诊断较为困难。术前可采用病理活检, 吸取部分骨腔内容物进行检查, 判断内容物的成分, 与成釉细胞瘤和牙源性角化囊肿等颌骨肿物相鉴别, 以辅助疾病诊

断^[24-25]。SBC组织病理学检查^[26]结果显示:疏松结缔组织覆盖骨壁,其中包含充血的毛细血管和渗出的血细胞、血黄素及多核巨细胞,术后病理诊断仍然是确诊SBC的重要指标。

由于SBC较为罕见,目前尚缺乏足够的技术支持,对SBC的治疗尚无公认的金标准。临床上常用的治疗方法包括刮治术、开窗术和保守治疗。本文作者认为刮治术是目前值得推荐的治疗方法。刮治术主要通过充分搔刮骨腔,刮除骨壁上可能附着的薄层结缔组织,并使骨腔内出血和血凝块机化,从而形成肉芽组织,在成骨细胞和破骨细胞的作用下逐渐成骨,以达到骨再生修复骨腔的目的^[27]。此外,术前仅依靠影像学检查、病史采集及专科检查很难将SBC与其他类型颌骨囊肿鉴别,手术探查同时也可帮助确诊疾病,避免误诊。有学者^[10]认为手术中应进行自体骨或异体骨移植。但SBC好发于20岁以下,患者的身体机能较好,无需进行骨移植,术后定期复查观察病变范围,并行病变累及牙齿牙髓的电活力测试。关于SBC的治疗也可采用微创手术,利用内窥镜观察病变骨腔内情况,进而明确诊断,但术中需要搔刮骨腔,出血机化成骨^[28]。采用保守治疗方式的SBC患者,需严格遵医嘱,定期观察,防止病变范围扩大形成病理性骨折^[29]。对于病变范围较大的SBC患者,血凝块形成无法修复整个骨腔,可能使病变继续存在,可先采用颌骨肿物开窗术,术后复查确定病变的大小,决定是否需要进行二期手术^[28]。存在硬骨板缺失、扇形边缘、结节状骨质扩张和多个骨腔的SBC患者术后复发的可能性较高,应注意随访^[30]。该患者采用骨开窗后刮治术,使新鲜血充满骨腔中,有助于机化成骨,术后恢复良好,无复发及其他并发症。SBC预后良好,复发率较低,对于其预后可借助影像学资料预测患者术后复发情况^[10]。

本文作者报道了1例同时发生于同侧上下颌骨的SBC患者,较为罕见。SBC无特征性的临床和影像学表现,极易与颌骨常见疾病混淆,若密度减低区无边缘骨白线,应注意是否为颌骨SBC。治疗方式可首选刮治术,利用血凝块机化成骨,患者预后良好且复发率低。

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