

Basal cell adenocarcinoma of the maxillary sinus: A case report

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Case report

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Abstract

Background: A basal cell adenocarcinoma (BCAC) is a low-grade malignancy of the salivary glands. A BCAC of the minor salivary gland is a rare disease, which is extremely rare in the maxillary sinus without invading from the palate and buccal mucosa. The histopathological characteristics of a BCAC are similar to those of a basal cell adenoma (BCA). However, BCAC can be differentiated from BCA based on its tendency to invade surrounding tissues. Surgical resection is the first-line treatment for BCACs. We report a case of a BCAC arising from the maxillary sinus minor salivary glands in an 82-year-old man.

Case presentation: In 2016, the patient presented with recurrent epistaxis, and he was referred to our department because a tumor was found in his left nasal cavity. Gross resection using the Denker operation was performed. Histopathological examination revealed no surrounding tissue invasion; therefore, BCA was diagnosed.

In 2017, the recurrent lesion was resected using endoscopy as much as possible, and the histopathologic findings again revealed a BCA.

In 2019, he developed diplopia, frequent epistaxis, and buccal swelling. The recurrence of maxillary sinus tumor was shown again with invasion of surrounding tissues, and we presumed a clinical diagnosis of a left maxillary carcinoma (suspected BCAC). When we performed partial maxillary resection by the Weber–Ferguson incision, we found that the tumor had partially invaded the bone of the orbital floor; thus, the floor of the orbit and orbital fat were partially resected. Finally, the tumor was diagnosed as a BCAC.

Conclusion: We report a BCAC arising from the maxillary sinus. This is an extremely rare sinus tumor, and differentiation of BCAC and BCA is important from a clinical or histopathological examination. A partial maxillectomy with a Weber–Ferguson incision was required for complete resection. When a malignancy is suspected in the recurrent maxillary sinus tumor, it is important to have a wider surgical field than that of the previous surgery to ensure complete resection.

1. Background

A basal cell adenocarcinoma (BCAC) is a rare tumor of the salivary gland and most commonly affects the parotid gland. A BCAC of the major salivary gland was first reported by Ellis et al. [1], and since then only three cases have been reported with the development of BCACs in the maxillary sinus [2,3]. A BCAC often invades from palate and buccal mucosa to the maxillary sinus, and it is extremely rare to develop from the maxillary sinus independently, because minor salivary glands are most frequently recognized in the buccal mucosa and hard palate. Similarly, paranasal BCACs probably originate from minor salivary gland tissue, but the distribution of the minor salivary gland in the maxillary sinus is not known. BCAC is histologically similar to BCA, and differentiation can be especially challenging with small biopsy specimens. As per the fourth edition of the World Health Organization Classification of Tumors, BCACs are classified as low-grade adenocarcinomas with a favorable prognosis [4]. Minor salivary gland tumors

have a tendency of infiltrating surrounding tissues and therefore require wide local excision to ensure complete resection. Here, we report a case of a BCAC arising from the maxillary sinus.

2. Case Presentation

In 2016, an 82-year-old man visited an otolaryngology clinic with a complaint of recurrent epistaxis. Upon examination with a video fiberscope, a tumor was found in the left nasal cavity, and he was referred to the Department of Otorhinolaryngology–Head and Neck Surgery, Juntendo University Urayasu Hospital. He had a history of angina, cerebral infarction (for which oral aspirin was prescribed), and diabetes mellitus. The paranasal computed tomography (CT) scan revealed soft tissue shadow completely occupying the left maxillary sinus (Figure 1) and bone destruction of the orbital floor. The histopathological findings of outpatient biopsy showed a benign salivary gland tumor. Therefore, gross resection using the Denker operation was performed. Additional wide resection was not performed because histopathological findings showed no surrounding tissue invasion. At this point, the tumor was diagnosed as a BCA.

After six months, magnetic resonance imaging (MRI) performed for periodic inspection revealed recurrence in the orbital floor. As the patient was asymptomatic, a watch-and-wait approach was undertaken per the patient's request.

In 2017, the patient experienced epistaxis from the gradually growing tumor, and the recurrent lesion was resected using endoscopy as much as possible. The histopathologic findings again revealed a BCA. Subsequently, the tumor recurred and gradually increased in size.

In 2019, the patient developed diplopia, frequent epistaxis, and buccal swelling. A hemorrhagic tumor filling the left nasal cavity was observed. Further examination revealed no abnormalities of the oral mucosa, no adhesion of the tumor to the skin, no facial paralysis, and no cervical lymphadenopathy. Paranasal CT with contrast revealed a tumor in the left maxillary sinus (Figure 2a). The bone of the orbital floor was defected due to tumor invasion. Further, MRI findings showed subsequent sinusitis in the left ethmoid and frontal sinuses, suggesting no tumor infiltration (Figure 2b), and no infiltration of the skull base. Moreover, whole-body CT revealed no distant metastasis. Blood test results including the tumor marker, carcinoembryonic antigen level, showed no abnormalities.

Based on these findings, the clinical diagnosis was a left maxillary carcinoma (suspected BCAC) with a TNM Classification of Malignant Tumors of cT4aN0M0. Although the patient was elderly, his general condition was good; thus, we planned to resect surgically. He had requested to preserve his eye; therefore, we decided upon partial maxillary resection with as-needed postoperative radiotherapy. Partial maxillary resection was performed by Weber–Ferguson incision. The maxilla had not been infiltrated by the tumor and was reserved (Figure 3). The tumor had partially invaded the bone of the orbital floor; thus, the floor of the orbit and orbital fat were partially resected. We were able to preserve the inferior rectus muscle. After excision of the tumor, skin grafting was performed on the anterior wall of the maxillary sinus to prevent scar contracture.

The tissue had a palisading pattern similar to a BCA (Figure 4a); nonetheless, some venous and bone infiltration was observed (Figure 4b). Based on the above observations, we diagnosed the tumor as malignant. The tumor showed a solid-type structure (Figure 4c) and was very fragile, which made it difficult to judge the surgical margin. In addition to the markers such as Ki-67, p53, epidermal growth factor receptor (EGFR), and B-cell lymphoma-2 (*bcl-2*), protein staining was difficult to evaluate due to the decalcification procedure. Retrospectively we performed immunohistochemical analysis for the first surgical sections; the tumor cells were only immunoreactive with EGFR (Figure 5).

The patient was discharged on day 21 postoperatively without any major postoperative complications. His diplopia improved as the wound stabilized. As per the patient's request, the treatment strategy was followed up without postoperative radiotherapy. No recurrence was found on the CT scan obtained six months after surgery, and he has had a good clinical course (Figure 6).

3. Discussion And Conclusions

The incidence of malignant salivary gland tumors is approximately $\leq 3\%$ [5], with around 90% of such tumors developing in the parotid gland [1,5]. The incidence of palatal BCACs as malignant tumors in minor salivary glands ranges from 0.26% [6] to 1.2% [7]. A BCAC commonly occurs in people who are approximately 60–70 years old with similar frequency in males and females [1]. Cuthbertson et al. reviewed 72 cases of BCACs arising in the minor salivary glands and reported that the palate was the most common location with a high local recurrence rate (41%). Out of the 72 cases, only seven had BCACs localized in the nasal cavity and sinuses [8]. To our knowledge only three cases have been reported so far on BCACs arising from the maxillary sinus [2,3]; thus, a BCAC in the maxillary sinus is an extremely rare disease.

There are two types of histopathologic findings in BCACs: 1) small, round cells with basophilic nuclei and lack of cytoplasm and 2) slightly larger, multinucleated cells with eosinophilic cytoplasm and pale-staining basophilic nuclei. A palisading pattern is observed in the tumors, and the tissue structure of these tumors is similar to that of benign BCAs. Depending on the dominant tumor patterns found in the tissue, tumors are structurally classified into solid, tubular, trabecular, or membranous [1]. The solid type is the most common and was observed in this case. The possible differential diagnoses include BCA, adenoid cystic carcinoma, small cell carcinoma, and metastatic basal cell carcinoma with the primary tumor originating from the skin. The differentiation of a BCAC from a BCA is based on invasive proliferation into nerves, blood vessels, and surrounding tissues, necrosis, mitotic figures, and anisonucleosis [4,9]. Invasive proliferation into nerves and blood vessels is seen in 25–35% of BCAC cases [10]. Both BCACs and BCAs differentiate into myoepithelial cells, which makes it impossible to differentiate these tumors based on immunohistochemical findings alone. However, Nagao et al [5] reported that a higher rate of cell proliferation (> 4 mitotic counts/10 high power field (HPF) or 5% of the Ki-67 labeling index) and apoptosis, along with increased expression of p53 and EGFR and a loss of *bcl-2* expression are markers for diagnosis of BCACs. In this case, the specimen of the primary operation was diagnosed with a benign tumor without having the invasive tendency to a surrounding tissue.

Retrospectively we performed immunohistochemical analysis for this specimen; the tumor cells were only immunoreactive with EGFR. Ki-67, p53 and *bcl2* were negative, but it was necessary for us to think about a malignant tumor at the first operation. With the third surgery, the tumor originated in the sinus cavity, and since a large amount of bone was attached to the tumor specimen, decalcification was performed prior to immunohistochemical staining. Therefore, the evaluation of Ki-67, p53, EGFR, and *bcl-2* staining was difficult, and ultimately, the most important diagnostic factor for distinguishing BCAC from BCA was the infiltrative growth pattern.

Wide local resection with confirmation of negative margins has been recommend as the first choice for BCAC of maxillary sinus. However, there have been reports of radiotherapy in cases where surgery was not an option owing to reduced performance status or other reasons. Successful local control and quality of life maintenance have been reported with radiotherapy alone [11]. It may not be possible to make a correct diagnosis of small salivary gland tumors originating from the paranasal sinuses with biopsy alone; hence, treatment may require multiple operations, as in this case. Therefore, it is important to ensure that the tumor is completely resected with a good operating field, because malignant transformation may have occurred since the initial surgery.

Although based on the histopathology, a BCAC is classified as a low-grade adenocarcinoma, a BCAC arising in minor salivary glands is reported to have local recurrence, distant metastasis, and mortality rates of 71%, 21%, and 29%, respectively [10]. In the present case, the BCAC developed in the maxillary sinus, which made it difficult to obtain an adequate surgical margin. Originally, it was considered preferable to also perform postoperative radiotherapy, but the patient declined this treatment. Therefore, the patient's progress was being actively monitored, which required careful follow-up.

In conclusion, we report a rare case of a BCAC arising from maxillary sinus minor salivary glands. This case is a perfect example of the difficulties in pathologic differentiation, in which this tumor was twice diagnosed a BCA before a BCAC was rendered. A partial maxillectomy with Weber–Ferguson incision was required for complete resection. When a malignancy is suspected in a recurrent maxillary sinus tumor, it is important to have a better surgical field than that of the previous surgery to ensure complete resection.

Declarations

Not applicable

Ethics approval and consent to participate

Not applicable

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Availability of data and materials

Not applicable

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

MK and SI contributed equally to this work. MT, KS and AY conceived of the study and participated in its design and coordination. KI drafted the manuscript. SI and RH were involved in revising the manuscript. All authors read and approved the final manuscript.

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Not applicable

Abbreviations

BCA, basal cell adenoma; BCAC, basal cell adenocarcinoma; *bcl-2*, B-cell lymphoma-2; CT: computed tomography; EGFR, epidermal growth factor receptor; MRI, magnetic resonance imaging, HPF, high power field

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Figures

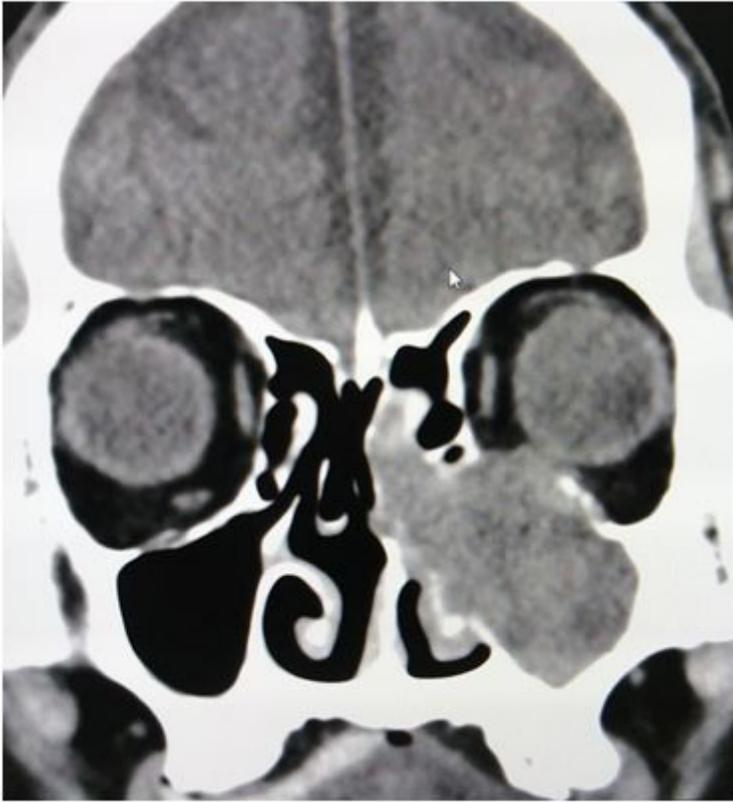


Figure 1

Initial paranasal CT findings (coronal section): Soft tissue density occupied left maxillary sinus and infiltrated in the orbit floor.

Figure 2 (a)



Figure 2 (b)

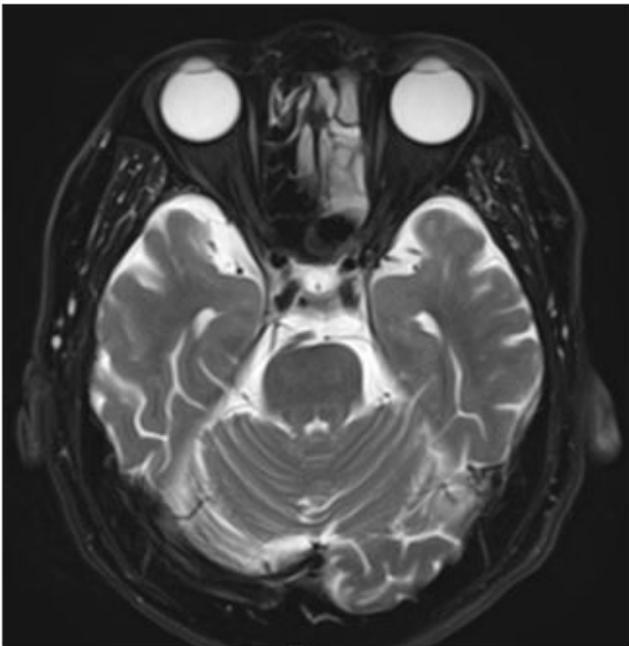


Figure 2

Paranasal CT and MRI findings at recurrence: (a) CT (coronal section) with contrast effect showed the tumor in the left maxillary sinus, bone destruction of the orbital floor, and invasion of the anterior part of the orbital contents. (b) MRI with T2-weighted image (axial section) showed subsequent sinusitis in the frontal sinus and superior part of the ethmoid sinus.

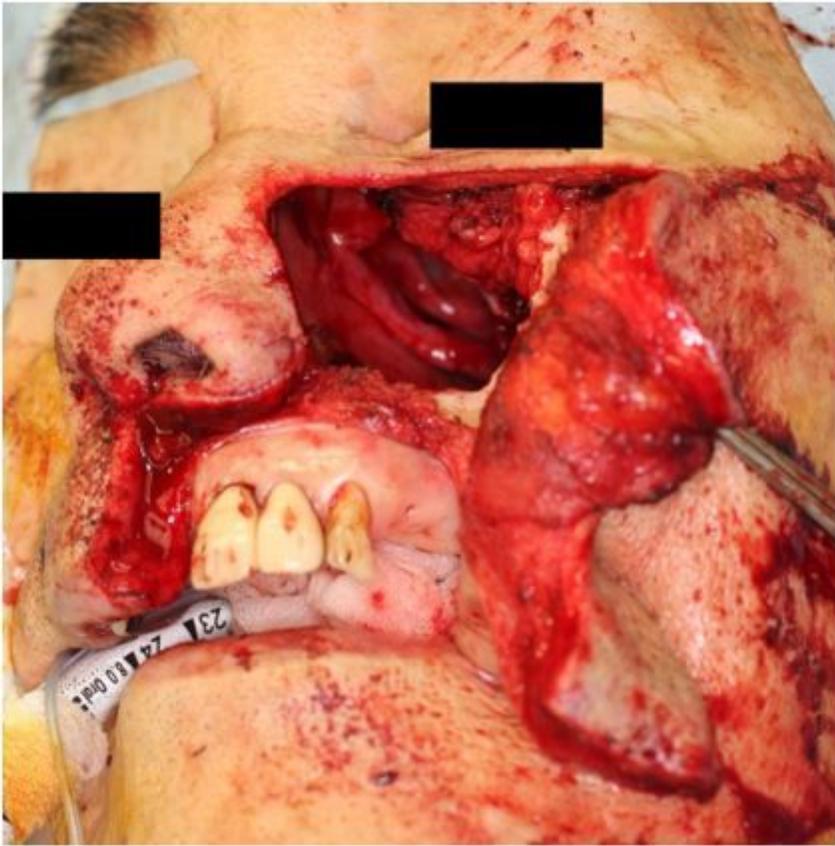


Figure 3

Operative findings: A partial maxillary resection using Weber–Ferguson incision is shown. The tumor had partially infiltrated the bone of the orbital floor, so the floor of the orbit and orbital fat were partially resected.

Figure 4 (a)

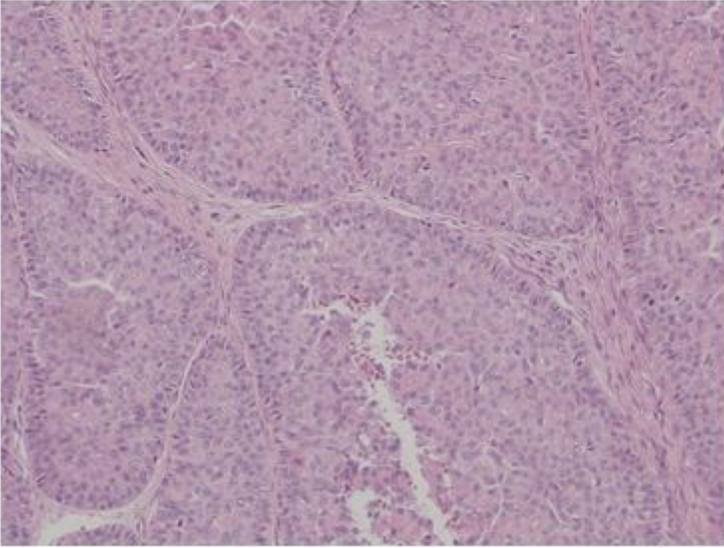


Figure 4 (b)

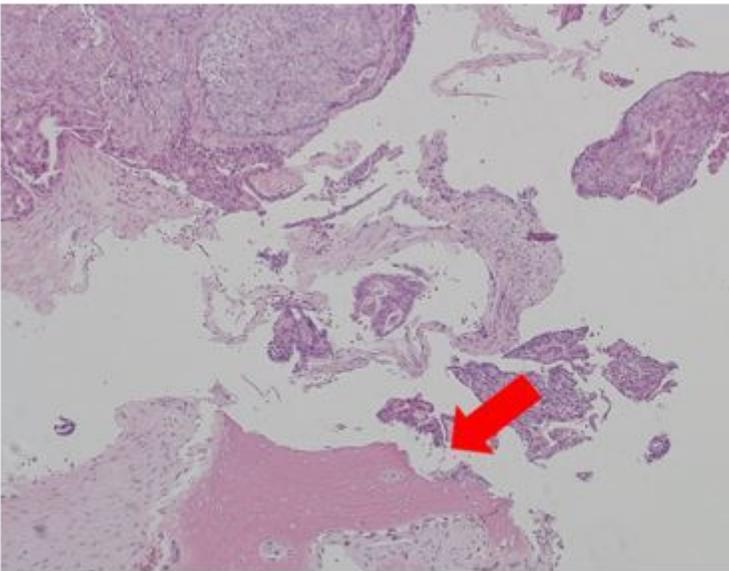


Figure 4

Pathologic findings: (a) A tissue sample showed a palisading pattern similar to that of a basal cell adenoma. (b) The red arrow shows bone infiltration of the tumor, so the tumor was considered to have undergone malignant transformation. (c) Most of the tumor showed solid growth, indicating a solid-type growth pattern.

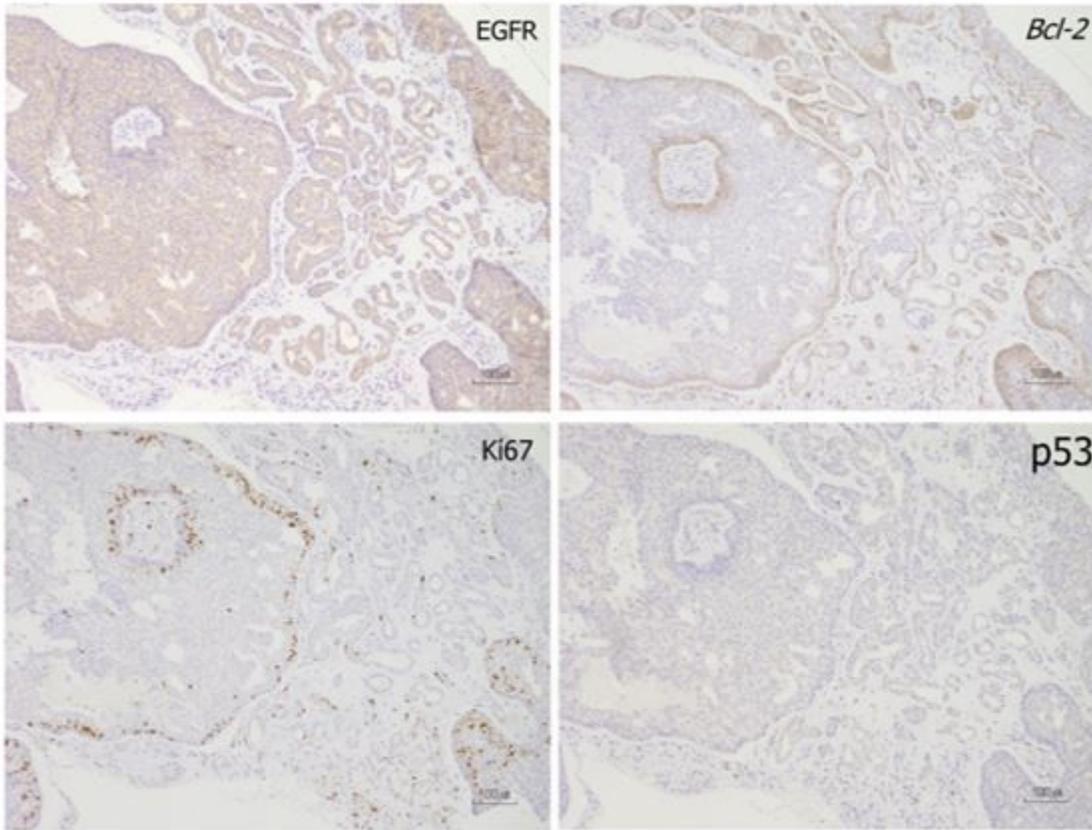


Figure 5

Immunohistochemical findings. The tumor cells were only immunoreactive with EGFR. Ki-67, p53 and bcl2 were negative.

Figure 6 (a)

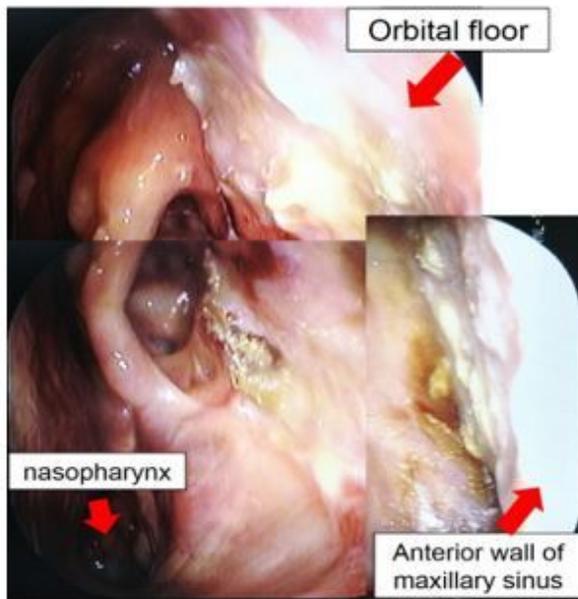


Figure 6 (b)



Figure 6

Post-operative endoscopic and CT findings: (a) Intranasal findings six months postoperatively showed good epithelialization of the grafted skin and no tumor recurrence. (b) CT obtained six months postoperatively showed no findings suggestive of recurrence.