Abstract

Basaloid squamous cell carcinoma (BSCC) is a histologically distinctive variant of squamous cell carcinoma comprising basal cell carcinoma and squamous cell carcinoma. BSCC is aggressive and shows a poor prognosis because of frequent lymph node invasion and distant metastases. BSCC preferentially occurs in the cervix, thymus, and esophagus and is uncommonly found in the head and neck region. BSCC in the nasal cavity or paranasal sinus is particularly rare. Inverted papilloma is an uncommon, benign tumor with a propensity to be associated with malignancy; however, BSCC arising in an inverted papilloma has never been reported. Here we report a case of a 56-year old woman with BSCC arising in an inverted papilloma in the nasal cavity.

The woman was referred to our hospital for epistaxis, nasal congestion, and dysphagia. A tumor was observed to completely occupy the left nasal cavity. The biopsy specimen was histologically diagnosed as papilloma. Computed tomography demonstrated a tumor with heterogeneous contrast effect occupying the left nasal cavity; however, extra-nasal tract extension was not observed. We performed endoscopic excision of the tumor. Microscopic findings confirmed the diagnosis of BSCC arising from an inverted papilloma. No tumor recurrence has been observed for 13 months after surgery.

Key words: Basaloid squamous cell carcinoma, inverted papilloma, nasal cavity, head and neck, endoscopic surgery

Introduction

Basaloid squamous cell carcinoma (BSCC) is a histologically distinctive variant of squamous cell carcinoma comprising two parts: basal cell carcinoma and squamous cell carcinoma [1]. BSCC is aggressive and shows a poor prognosis because of frequent lymph node invasion and distant metastases. BSCC preferentially occurs in the cervix, thymus, and esophagus and is uncommonly found within the head and neck region. BSCC in the nasal cavity is especially rare; only 18 cases have been reported in the English literature [2-7].

Inverted papilloma (IP) is an uncommon benign tumor with a propensity to be associated with malignancy; however, BSCC arising in IP has never been reported in the English literature. Here we report a case of a patient with BSCC arising from IP in the nasal cavity.

Case

A 56-year-old woman was referred to our hospital with complaints of epistaxis, nasal congestion, and dysphagia. On endoscopic examination, a tumor was observed to completely occupy the left nasal cavity. A biopsy was performed, and the specimen was histologically diagnosed as exophytic papilloma. Computed tomography demonstrated a tumor with heterogeneous contrast effect occupying the left nasal cavity; however, extra-nasal tract extension was not observed (Fig. 1). A T1 stage papilloma was diagnosed according to Krouse classification, and definitive endoscopic excision was performed.

The operation was performed under general anesthesia. The tumor originated from the nasal septum in the left nasal cavity without any invasion toward the surrounding components (Fig. 2A). The tumor was resected at the root of the tumor and the tumor basal mucosa was cauterized by monopolar electrocautery (Fig. 2B).

Postoperative microscopic findings with hematoxylin and eosin staining revealed atypical cells with

hyperchromatic nuclei in scant cytoplasm spread out in inverted papilloma and composed of multiple variably sized and rounded nodules (Fig. 3A). The nodules of the tumor showed a peripheral palisade of atypical cells (Fig. 3B). On immunohistochemical study, cytokelatin 5/6 (Fig. 3C), a marker of squamous epithelial cells, and p63 (Fig. 3D), a marker of basal cells, were positive, respectively. MIB-1 index was calculated by Ki-67 staining (Fig 3E) and was 35%. These postoperative histological findings confirmed the diagnosis of BSCC arising in an inverted papilloma.

After surgery, ultrasonography and positron emission tomography/computed tomography were performed for the detection of cervical lymph node invasion and distant metastases; however, metastatic regions were not observed.

Endoscopic sinus surgery was performed for additional resection of the tumor basal mucosa, perichondrium of the nasal septum, and periosteum on the bottom of the nasal cavity. These resected tissues were examined microscopically, and no evidence of papilloma or malignancy was observed. We considered BSCC in IP to be completely resected with negative surgical margins; thus, additional treatments such as radiotherapy or chemotherapy were not performed. No tumor recurrence has been detected in the 13 months since the first surgery.

Discussion

Papilloma arising in the sinonasal tract is classified into three groups: IP, exophytic papilloma, and cylindrical cell papilloma. IP has a high incidence of recurrence and has a propensity to be associated with malignancy; it has been reported as approximately 11% of all papilloma. IP preferentially occurs at ethmoid and maxillary sinus (71% and 22%, respectively); however, it occurs at the nasal septum only 1% of the time. Preoperative pathological diagnosis of malignancy in IP is difficult due to its rarity, and accurate preoperative pathological diagnosis is also difficult because IP has two growth patterns: inverted or mixed inverted and exophytic [8]. Magnetic resonance imaging is useful in

evaluating the primary tumor extension, but it is difficult to distinguish malignancy in IP because of a significant overlap of imaging features [9]. Krouse indicated that the treatment of IP is characterized by the extent and location of the tumor [10]. To date, several studies have demonstrated the utility of an endoscopic approach for IP [11-13]. These articles indicated that endoscopic surgery for IP should be limited in cases of T1, T2, and selected T3 tumors [13, 14]. In our case, the tumor originated from the nasal septum and was limited to the nasal cavity, and no invasion toward surrounding tissues was observed. Therefore, the tumor was classified into the T1 stage according to the Krouse classification, and endoscopic surgery was selected as the most feasible treatment. However, because the preoperative diagnosis is different from the postoperative diagnosis, we should consider the possibility of other papilloma subtypes or synchronous malignancy and carefully treat to prevent recurrence.

Surgical margin is widely believed to be a significant prognostic factor in head and neck cancer, and it is also important for the treatment of cancers in the nasal cavity. Endoscopic surgery for sinonasal cancer has a limitation because of the difficulty in maintaining sufficient surgical margin; therefore, we should carefully choose a feasible approach. In our case, the tumor was in the early stage and pedunculated; moreover, it did not show macroscopic invasion toward surrounding components in the first surgery. In addition, nasal septum could be clearly observed and recurrence could be easily detected. In such a selected case, minimum surgical margin may be one of the feasible strategies to preserve nose function.

Adjuvant therapy for synchronous malignancy in IP is not yet well established owing to its rarity. Pasquini et al. [13] reported three cases of T1 stage IP with malignancy treated by endoscopic surgery, which did not receive any adjuvant therapy; no recurrence was observed. Thorp et al. [14] reported one case with a similar clinical course. In such early stage cases, complete resection of the tumor is feasible; thus, adjuvant therapy might be unnecessary. Postoperative radiotherapy has been recommended for high-risk groups of head and neck carcinoma such as multiple nodal metastases,

positive margin, and extracapsular spread [15-17]; however, patients should be carefully selected due to its toxicity.

BSCC is a histologically distinctive variant of squamous cell carcinoma composed of two parts: basal cell carcinoma and squamous cell carcinoma [1]. BSCC in the nasal cavity is extremely rare, and only 18 cases have been reported to date [2-7]. This is the first report of BSCC arising in IP. BSCC shows an aggressive behavior and carries a poor prognosis, especially in cases with an MIB-1 index >50% [18]; however, others have reported that long-term survival of BSCC in the sinonasal tract was equal to squamous cell carcinoma [19]. Ten cases of BSCC in the nasal cavity with T1 stage were reported, and all cases received surgical treatment; however, only three cases received postoperative radiotherapy [2-5]. These reports showed similar survival outcome regardless of the postoperative radiotherapy. Thus, adjuvant radiotherapy for synchronous malignancy in IP, if it was BSCC, might be considered carefully. In our case, the MIB-1 index was only 35% and the tumor was early stage, completely resected, and originated from the nasal septum, and thus, it could be clearly observed and recurrence could be easily detected. In such a selected case, observation only might be the preferred strategy; however, long-term follow-up is necessary.

Conclusion

We report the first case of BSCC arising in IP, which was classified as T1 according to Krouse classification and treated with endoscopic surgery. Treatment for synchronous malignancies in I has not been well established; however, adjuvant radiotherapy might be unnecessary for early stage and completely resected tumors.

Acknowledgment

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Conflict of interest

We have no conflict of interest in this case report.

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Figure 1 Computed tomography with contrast effect

Computed tomography showed a tumor occupying the left nasal cavity; however, the tumor was limited to the nasal cavity. Invasion toward surrounding components such as bone defects was not observed.

Figure 2 Intraoperative findings in the left nasal cavity

A) Before excision: Endoscopic surgery was performed and showed the tumor originated from the nasal septum. The tumor did not invade surrounding components, such as the inferior or middle nasal concha or bottom of the nasal cavity.
B) After resection: The tumor was completely resected and the tumor basal mucosa was cauterized by monopolar electrocautery.

Figure 3 Postoperative histopathological findings

A) Hematoxylin and eosin (HE) (×100): atypical cells were spread out in inverted papilloma (IP). The white arrowhead shows atypical (malignant) cells area, and the yellow arrowhead shows IP.
B) HE (×400): Cells with hyperchromatic nuclei and scant cytoplasm composed of multiple variable nodules showing cribriform growth pattern. Peripheral basaloid cells were palisaded.
C) Immunohistochemistry (IHC) with cytokeratin 5/6 (×400): Cytokeratin 5/6 was positive in the greater part of the cytoplasm of malignant cells.
D) IHC with p63 (×400): p63 was positively stained in the greater part of the nuclei of malignant cells.
E) IHC with Ki-67 (×400): MIB-1 index was calculated by Ki-67 staining and was 35%.

Figure 1

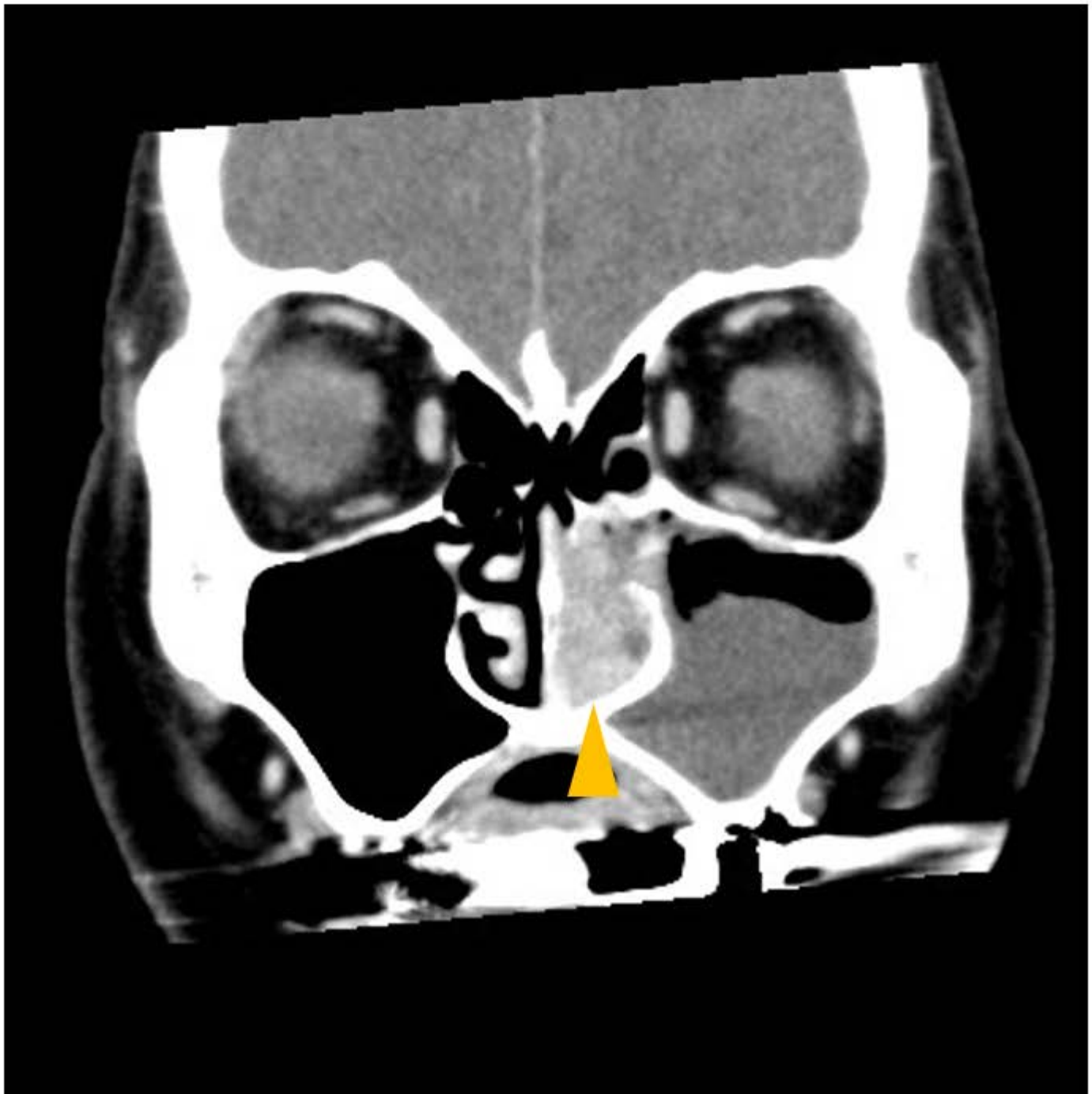
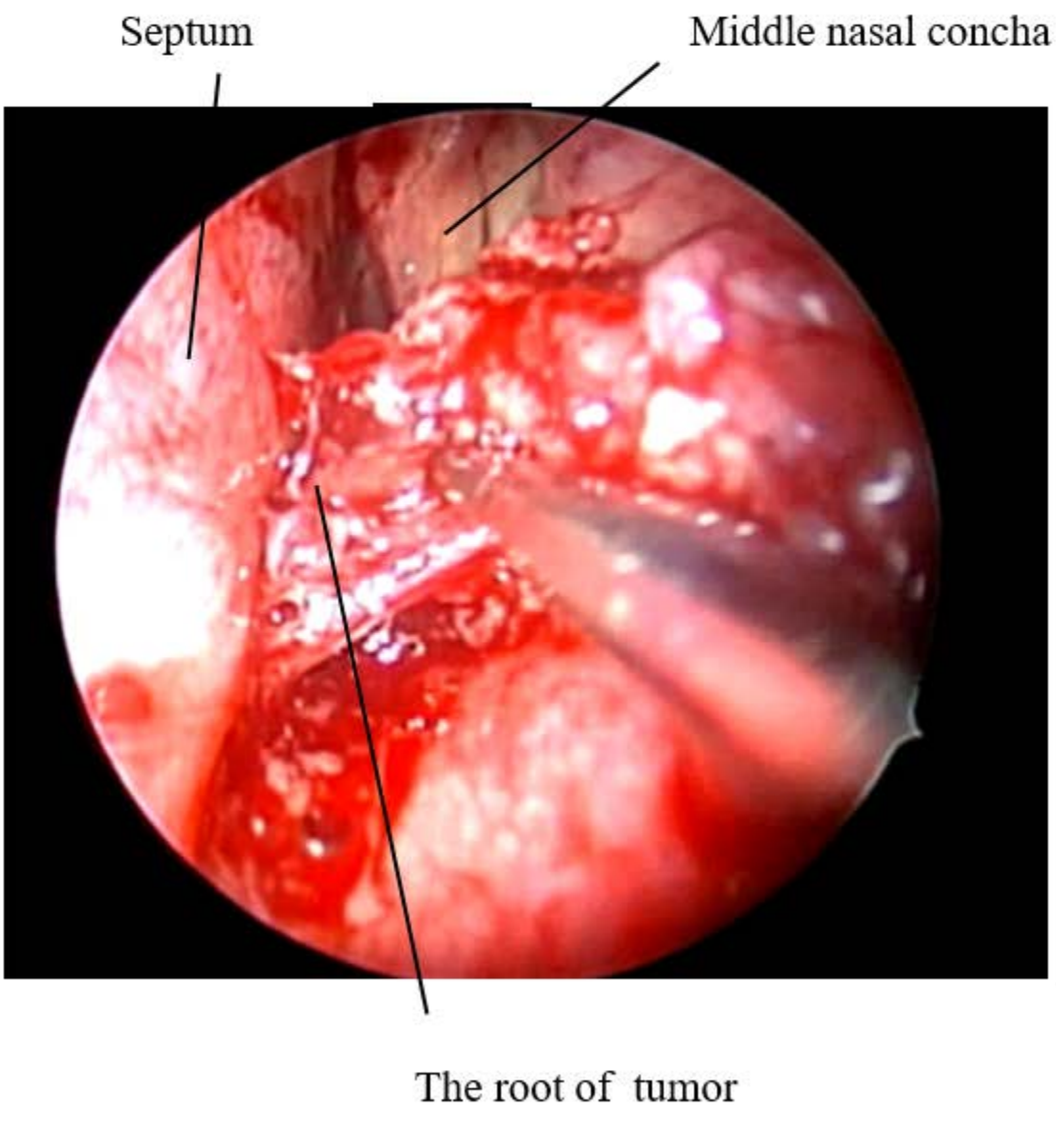
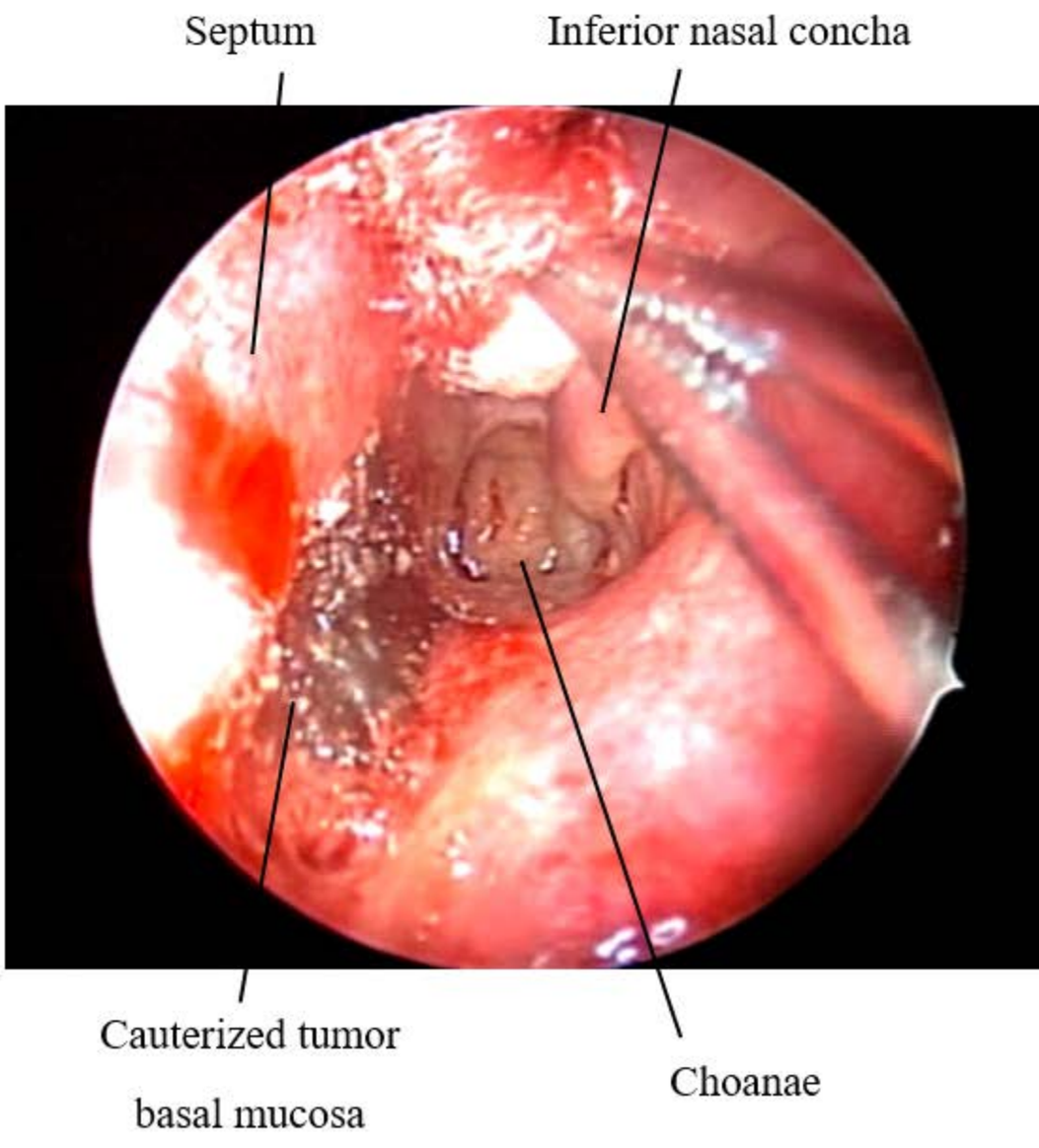


Figure 2

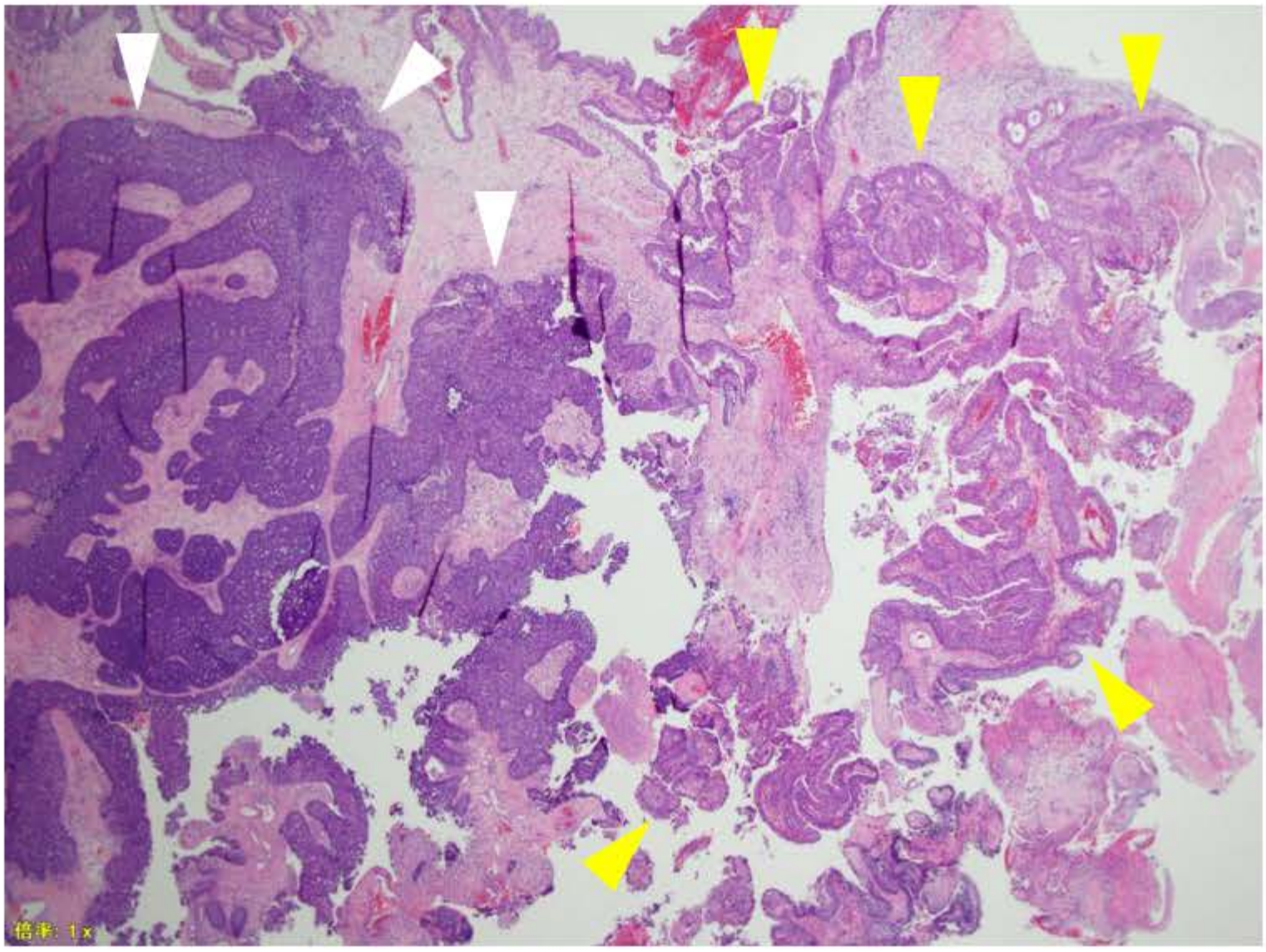


A) Before excision

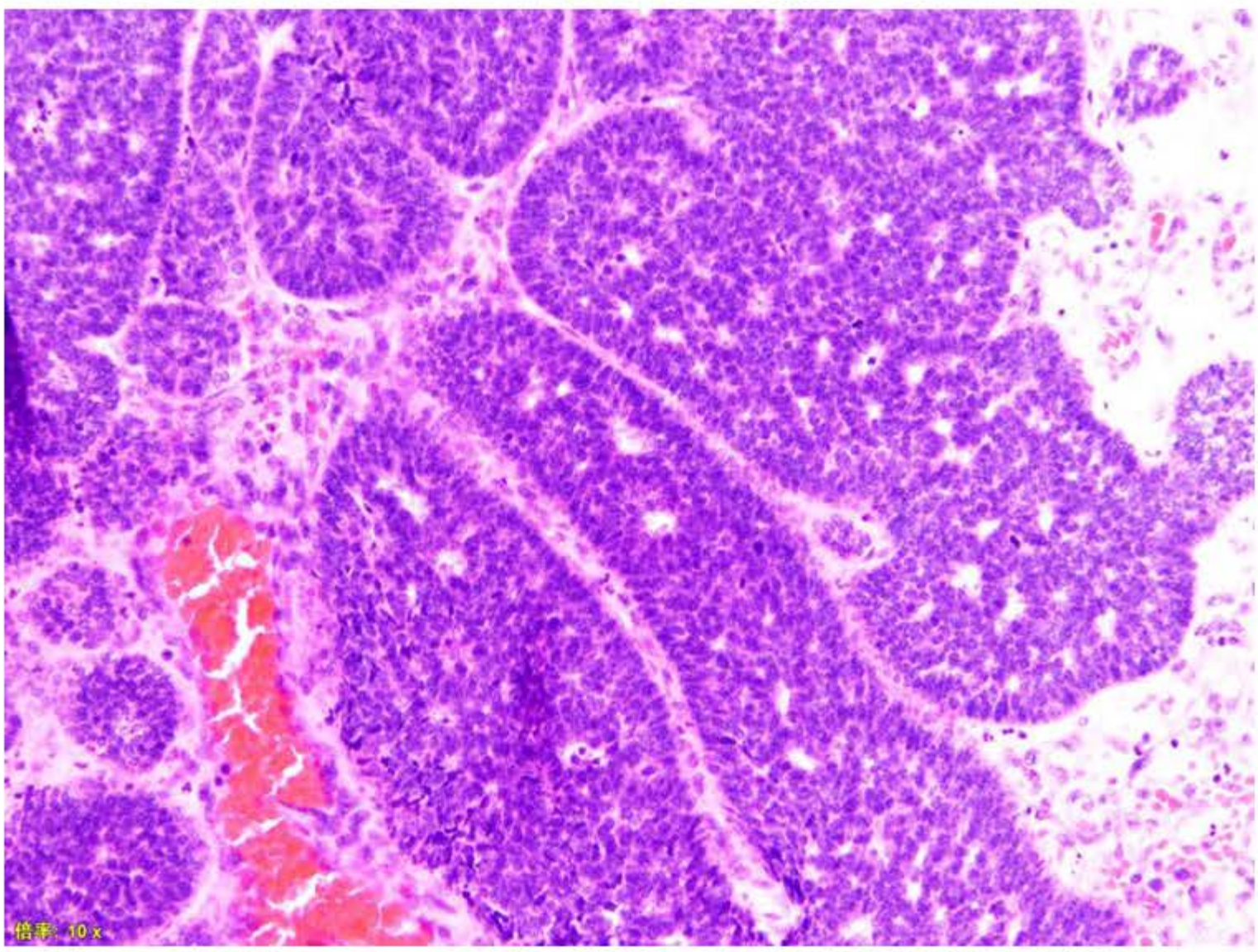


B) After resection

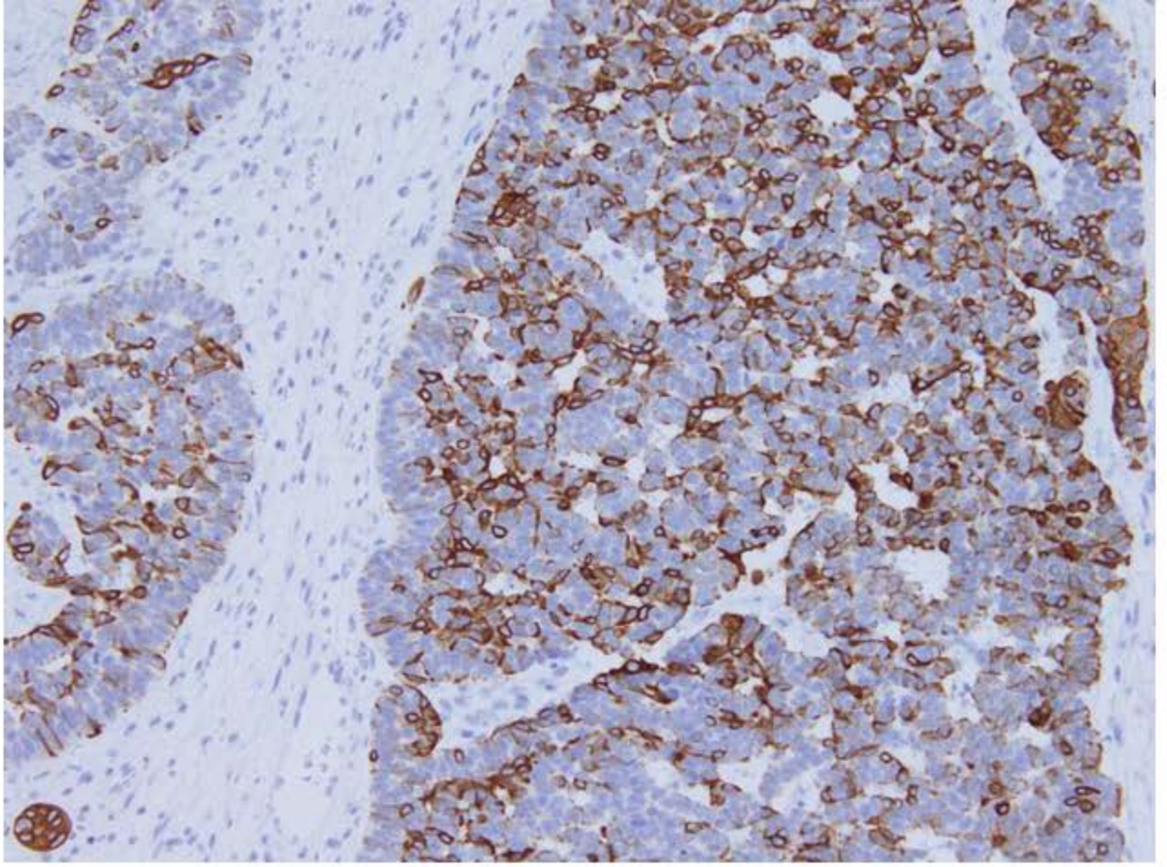
Figure 3



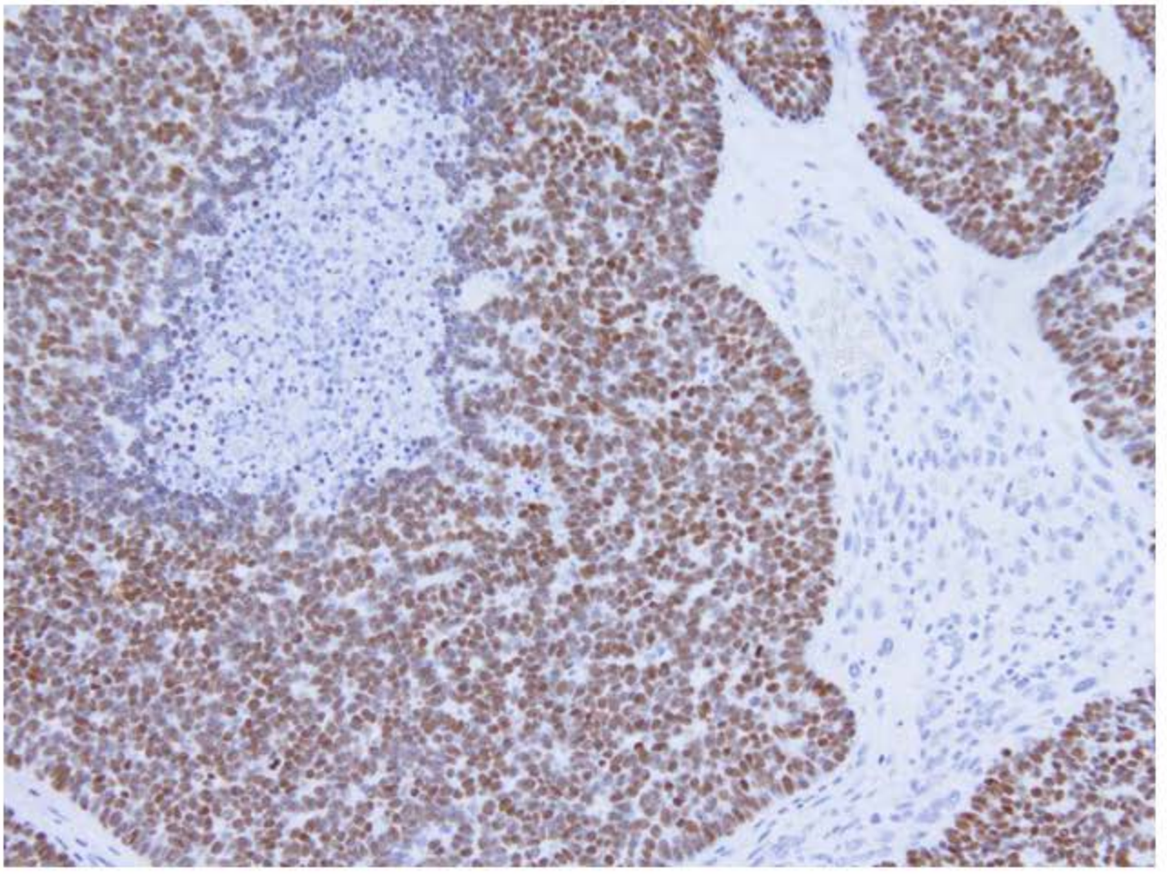
A) HE staining (low power field ×100)



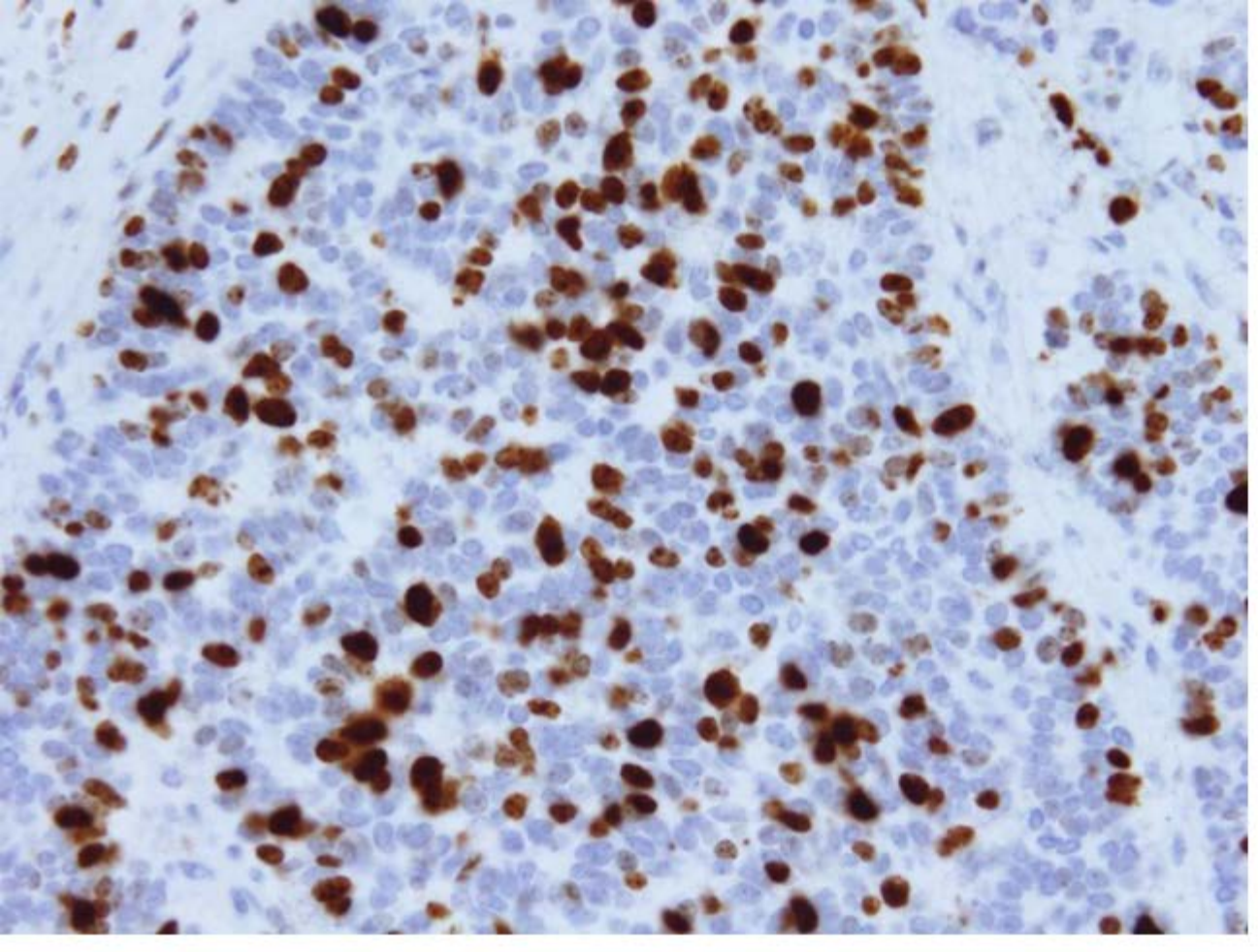
B) HE staining (high power field ×400)



C) Immunohistochemical staining with cytokeratin 5/6



D) Immunohistochemical staining with P63



E) Immunohistochemical staining with Ki-67

table 1.

Case No.	Age	Gender	Location	T Stage	Surgery	Preoperative RT	Postoperative RT	Preoperative CT	Postopetarive CT	Follow-Up (month)	Outcome	Reference
1	58	F	nasal cavity	1	Yes	No	No	No	No	17	NED	2
2	53	F	nasal cavity	1	Yes	No	Yes	No	Yes	96	STD	3
3	81	F	nasal cavity	1	Yes	No	No	No	No	36	NED	3
4	41	F	nasal cavity	1	Yes	No	No	No	No	60	AED	3
5	64	M	nasal cavity	1	Yes	No	No	No	No	48	NED	3
6	56	M	nasal cavity	1	Yes	No	Yes	No	No	24	NED	3
7	46	M	nasal cavity	1	Yes	No	No	No	No	8	AWD	3
8	86	F	nasal cavity	1	No	No	No	No	No	6	STD	3
9	79	M	nasal cavity	1	Yes	No	Yes	No	No	1	NED	3
10	36	M	nasal cavity	1	Yes	No	No	No	No	18	AWD	4
11	56	F	nasal cavity	1	Yes	No	No	No	No	13	NED	Present case