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Image interpretation for squamous cell carcinoma of Stensen's duct

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ABSTRACT

We report herein a case of squamous cell carcinoma presumed to have arisen from the right Stensen's duct. The patient, a 62-year-old man, was referred to our hospital with swelling in the right cheek. Magnetic-resonance imaging (MRI), including contrast-enhanced MRI, and contrast-enhanced computed tomography (CECT) enabled diagnosis of a solitary mass in Stensen's duct. Fat-suppressed T₂-weighted imaging, in particular, demonstrated a mass-like lesion in the dilated Stensen's duct and obstructive parotitis, where the duct transitions into the parotid gland. Gadolinium-DTPA-enhanced T₁-weighted imaging demonstrated the mass-like lesion surrounded by signal hyperintense layer showing continuous transition from the thickened Stensen's duct wall, which was also hyperintense. CECT revealed peripheral annular enhancement surrounding the tumorous mass, with no enhancement of the duct wall itself, reflecting an increase in micro blood vessels in the stroma of the neoplasm. These image findings correlated well with subsequent histopathological findings. A mass with rim-enhancement and dilated Stensen's duct accompanied by parotitis and no salivary calculus may suggest a differential diagnosis of malignant tumor of Stensen's duct.

Keywords: Stensen's duct, squamous cell carcinoma, parotitis, computed tomography, magnetic resonance imaging

INTRODUCTION

Primary carcinomas arising in Stensen's duct, which include mucoepidermoid carcinoma, undifferentiated carcinoma, adenocarcinoma, adenoid cystic carcinoma and squamous cell carcinoma (SCC), are relatively rare. To the best of our knowledge, only 8 cases of SCC have been reported in the English-language literature.¹⁻⁸ Moreover, only 3 studies have investigated the imaging features of SCC, and these focused on computed tomography^{8,9}, magnetic resonance imaging^{8,10} and ultrasonography⁸.

This report presents a case of SCC presumed to have arisen from Stensen's duct. Image interpretation and the potential for diagnostic use of magnetic resonance imaging (MRI), including contrast-enhanced MRI, and contrast-enhanced computed tomography (CECT) are discussed, and images obtained are collated to pathohistological findings.

CASE HISTORY

The patient was a 62-year-old man who presented at our hospital with swelling in the right cheek. He had been aware of the swelling for approximately 2 months prior to being referred to our hospital, but had not sought medical treatment, as no other inflammatory symptoms such as redness or heat were present. One week before initial examination at our hospital, he had visited an internal medicine department, as the swelling had begun to increase in size, with accompanying tenderness. The patient was subsequently introduced to

our hospital. Past and family medical histories revealed nothing of note.

Initial examination of the face revealed slight asymmetry due to diffuse swelling from the right cheek to the region of the right parotid gland. An induration, which showed tenderness on palpation, was located on the buccal surface of the right masseter muscle. In the oral cavity, disturbance was noted in excretion of saliva from the parotid papilla. Panoramic radiography was performed on suspicion of salivary calculus, but no abnormalities were revealed. Expansion and washing of Stensen's duct were undertaken with a dilating bougie over a period of approximately 2 weeks, but secretion of saliva gradually worsened. A catheter was then placed in the duct for 1 week. Discharge of pus remained markedly high, despite administration of an antibiotic and an anti-inflammatory. At this point, we began to suspect a local soft tissue mass disease.

After removal of the catheter, MRI and computed tomography (CT) were performed to determine whether we were dealing with a tumorous lesion or an inflammatory lesion, as initial clinical findings had suggested an inflammatory lesion such as sialolithiasis, sialoadenitis or sialodochitis in the parotid gland or Stensen's duct. First, turbo spin-echo MRI was performed due to the efficacy of this modality in the diagnosis of soft tissue disease. Fat-suppressed T₂-weighted imaging demonstrated a mass-like lesion in the right Stensen's duct, where the duct transitions into the parotid gland, with partial signal hypointensity in the central region of the lesion (Fig. 1a, b). This mass-like lesion showed a

distinct boundary contiguous with the buccal surface of the right masseter muscle. Signal intensities of the right Stensen's duct itself and the parotid gland were also higher than those on the left side. On T₁-weighted imaging (Fig. 1c, d), signal intensity of the mass-like lesion was relatively homogeneous and low, presenting a higher signal than closed masseter muscle. Furthermore, although the expanding mass appeared to press against the surface of the masseter muscle, no indications of the mass protruding from the duct were seen. On Gadolinium-DTPA-enhanced T₁-weighted imaging (Fig. 1e, f), the center of the mass-like lesion displayed signal hypointensity, suggesting necrosis. The lesion was surrounded by very signal hyperintense layer showing continuous transition from the thickened Stensen's duct wall, which was also hyperintense. This image also showed a very high signal intensity for the parotid gland where the duct had expanded, suggesting tumor infiltration or the presence of a coexisting inflammation. One week later, CECT was also performed for qualitative diagnosis of the central region of the mass-like lesion and to help determine the relationship between the lesion and the wall of Stensen's duct (Fig. 2a,b). Although the duct itself was dilated, the wall of Stensen's duct itself was not emphasized, differing from results of Gadolinium-DTPA-enhanced T₁-weighted imaging. Furthermore, although the lesion showed strong rim-enhancement with a hypodense area in the center of the mass, the layer of rim-enhancement seemed separate from the duct wall. No cervical lymphadenopathy was observed.

The results strongly suggested a tumorous lesion in Stensen's duct, and fine needle aspiration biopsy (FNAB) was performed. Cytological examination revealed a class V malignancy with cellular atypia of keratinized squamous epithelial cells.

Resection was performed under a preoperative diagnosis of malignant tumor of Stensen's duct accompanied by obstructive duct inflammation and parotitis (Fig. 3). Intraoperative pathological inspection revealed SCC. The tumor showed a consistently solid growth pattern with high atypia characterized by pleomorphism, hyperchromic nuclei, conspicuously large nucleoli and extensive atypical mitoses. Keratinization and cancer pearl formation were also recognized in the mass, indicative of well-differentiated SCC arising in the excretory duct of the parotid gland (Fig. 4a-d).

Postoperative course of the patient has been favorable, and no marked changes have been observed.

DISCUSSION

Stensen's duct is derived from excretory duct reserve cells, and runs along the surface of the masseter muscle from the anterior parotid gland to the orifice, localizing at the oral vestibule of the maxillary second molar tooth. Primary carcinoma arising in Stensen's duct is extremely rare. To the best of our knowledge, only 20 cases, including SCC¹⁻⁸, mucoepidermoid carcinoma¹⁰⁻¹³, adenocarcinoma¹⁴, adenoid cystic carcinoma⁹ and

undifferentiated carcinoma^{15,16}, have been reported in the English-language literature. In addition to these cases, a histological group of ductal carcinomas comprising a variant of primary parotid gland carcinoma has also been reviewed as “Stensen’s duct carcinoma”.^{7,12} Furthermore, the term “salivary duct carcinoma” has been used to describe such unusual tumors arising within the substance of the parotid gland,¹⁷⁻²⁰ with these tumors sharing many of the histopathological characteristics of “ductal carcinomas”, such as intraductal mammary and prostatic carcinomas. Ductal carcinoma usually presents as a parotid gland swelling and arises from the larger ducts in the parotid gland. Stensen’s duct carcinoma presents as a mid-cheek swelling,⁶ a mass within the buccal sulcus⁴ or with intermittent swelling of the parotid gland due to duct obstruction²¹. In this way, the terms “ductal carcinoma” and “salivary duct carcinoma” are often confused with “Stensen’s duct carcinoma”. Whatever the case, SCC and mucoepidermoid carcinoma predominate in Stensen’s duct carcinoma, with 8 and 6 cases reported, respectively.

Only 3 case reports describing SCC, adenoid cystic carcinoma and mucoepidermoid carcinoma have mentioned image findings for Stensen’s duct carcinoma, and these have focused on the imaging features of CT^{8,9}, MRI^{8,10} and ultrasonography⁸. The dates of these papers, appearing since 1980, may strongly reflect the spread of each imaging modality. Before these imaging modalities came into widespread use, correct diagnosis of carcinoma was delayed,^{5,6} with the condition often misdiagnosed as parotitis with ductal calculus or

recurrent parotitis.²¹ Traditional radiographic sialography is rarely useful owing to duct obstruction, and has been reported to miss the presence of a tumorous mass in Stensen's duct.^{7,12} Magnetic resonance sialography with heavily T₂-weighted images has recently come to be considered a new and promising alternative to traditional radiographic sialography.^{10,22,23} Sialoendoscopy may also prove reliable for obtaining information on salivary duct diseases as the technology develops.^{10,24} MRI and CT are currently the most commonly employed modalities in clinical practice for obtaining preoperative diagnosis of mass lesions of Stensen's duct.

In the present case, a combination of MRI and CECT proved effective in obtaining a preoperative diagnosis, providing correct and definitive positional information on the tumorous lesion. Based on previous evidence, the most prevalent site for primary carcinoma arising in Stensen's duct is just proximal to the orifice. A case report by Tominaga *et al.*⁸ noted SCC arising from the terminal end of Stensen's duct, close to the orifice. This was similar to the findings in another case described by Carpenter *et al.*⁹. In the present case, however, the mass-like lesion was located at the outside superior margin of the masseter muscle, bordering on the junction of the secretory portion of the parotid gland.

Using a combination of T₁- and T₂-weighted imaging, the possibilities of retention cyst, masseteric abscess, skin appendage tumor and swelling of a buccal lymph node were easily eliminated, due to the location of the lesion limited within the dilated Stensen's duct.

We were also able to rule out swelling of the accessory parotid gland. Tumors arising from the accessory parotid gland are not usually associated with obstruction of Stensen's duct.²⁵ The signal intensity of the mass-like lesion itself also ruled out a calcification lesion, although we were unable to completely exclude the possibility of a noncalcified stone. A noncalcified stone in Stensen's duct may appear as a low or middle signal lesion within a dilated, obstructed, saliva-filled duct. In the present case, T₂-weighted imaging showed signal hyperintensity in the secretory portion of Stensen's duct, with a markedly dilated and thickened, signal hyperintense wall. These findings may suggest inflammation in the duct and a disorder of salivary debouchement in the secretory portion, that is, sialodochitis caused by a mass-like lesion such as a noncalcified stone. Determining whether the mass-like lesion found in Stensen's duct in this case was indeed a neoplastic mass was thus important.

Contrast-enhanced T₁-weighted imaging also depicted a thickened wall with high signal intensity, in addition to the contrast enhancement effect observed in the center of the mass-like lesion. Such a signal intensity in a mass-like lesion suggests a parenchymal mass, that is to say, a neoplastic, tumorous mass lesion. Although these findings resembled those of a case reported by Tominaga *et al.*⁸, our contrast-enhanced findings showed that the contrast-enhanced thickened wall had avoided the tumorous mass altogether. In other words, the layer of rim-enhancement surrounding the mass represented a continuous

transition from the thickened duct wall. This made us consider the possibility that the contrast-enhancement surrounding the mass simply represented the thickened wall of Stensen's duct. Usually, with CECT, a contrast-enhanced image of a neoplastic mass such as a malignant tumor originating in soft tissue display heterogeneous high or middle density with an unclear rim-enhancement effect. In a lesion involving intratumoral necrosis, a low-density lesion with unclear rim-enhancement will be evident. If a mass-like lesion represents a neoplastic tumorous mass, the rim-enhancement effect should typically completely surround the lesion. In fact, CECT in a case reported by Carpenter *et al.*⁹ described a typical image of a neoplastic tumorous mass with completed rim-enhancement surrounding the mass. In the present case also, with CECT, an unclear peripheral rim-enhancement was only seen at the circumference of the tumorous mass. Stensen's duct wall was not enhanced and was not differentiated from the appearance of the duct, unlike in contrast-enhanced T₁-weighted imaging. Furthermore, CECT for sialodochitis caused by a noncalcified stone usually reveals a clear thickening and clear enhancement of Stensen's duct wall with low attenuation in the duct, unlike in our case.²⁶ Consequently, this localization convinced us that the lesion represented a tumor. Subsequent immunohistochemistry after enucleation of the tumor revealed vascular hyperplasia around the tumor (Fig. 5). Peripheral annular enhancement on CECT resulted from fibrous connective tissue with vascular hyperplasia around the tumor, while middle-density areas in the center of the

mass suggested a tumor accompanied by a large quantity of keratinized matter.

When a malignant tumor develops in Stensen's duct, inflammation may occur within the duct itself. In the differentiation of secondary inflammatory diseases caused by malignant neoplastic lesions or other lesions, salivary stagnation and a dilated excretory duct complicate the diagnosis of underlying abnormalities of Stensen's duct by magnetic resonance signal intensity, as the salivary excretory duct is an extremely narrow apparatus compared with other soft tissues, and the contrast resolution of MRI is superior to CT. Determination of whether the rim-enhancement around the mass lesion reflects vascular hyperplasia associated with tumor development or merely thickening of the duct walls due to inflammation may be crucial for estimating primary tumors occurring in Stensen's duct. A combination of MRI including contrast-enhanced T₁-weighted imaging and CECT may be necessary to obtain a more accurate diagnosis and localization of the tumor, even though MRI alone may be sufficient to indicate a tumorous mass with localized swelling along Stensen's duct or obstructive parotitis.

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FIGURE LEGENDS

Figure 1: a,b) Fat-suppressed T₂-weighted imaging.

*Mass-like lesion surrounded by a signal hyperintense rim shows continuous transition from the signal hyperintense wall of Stensen's duct (arrow).

c, d) T₁-weighted imaging.

*Mass-like lesion appears to press against the surface of the masseter muscle (arrows).

e, f) Contrast-enhanced T₁-weighted imaging.

*The very signal hyperintense rim surrounding the mass-like lesion clearly shows continuous transition from the hyperintense wall of Stensen's duct (arrows) on contrast-enhanced imaging.

Figure 2: a, b) Contrast-enhanced CT

*Stensen's duct wall itself is not enhanced. An unclear peripheral rim-enhancement is apparent only at the circumference of the mass (arrows).

Figure 3: Macroscopic findings (cut surface) following surgical resection.

Figure 4: Histopathological findings with HE staining

a) Microscopically, tumor masses were recognized at Stensen's duct in the area of the

parotid gland.

b) Dilated Stensen's duct was mostly occupied by tumor cells and tumor was encapsulated by partially fibrotic connective tissue.

c) Tumor was contiguous with ductal lining epithelium. Ductal lining epithelium showed various types of epithelial dysplasia, from moderate to severe or carcinoma in situ.

d) Tumor cells showed a high degree of atypia, including pleomorphism, hyperchromatic nucleus, conspicuous large nucleolus, and numerous atypical mitosis. Tumor cell invasion into peripheral fibrous connective tissue was observed focally, but no direct invasion of parotid gland lobules was recognized.

Figure 5: Immunostaining shows α -smooth muscle actin in ambient stroma of the neoplasm.

Fig.1

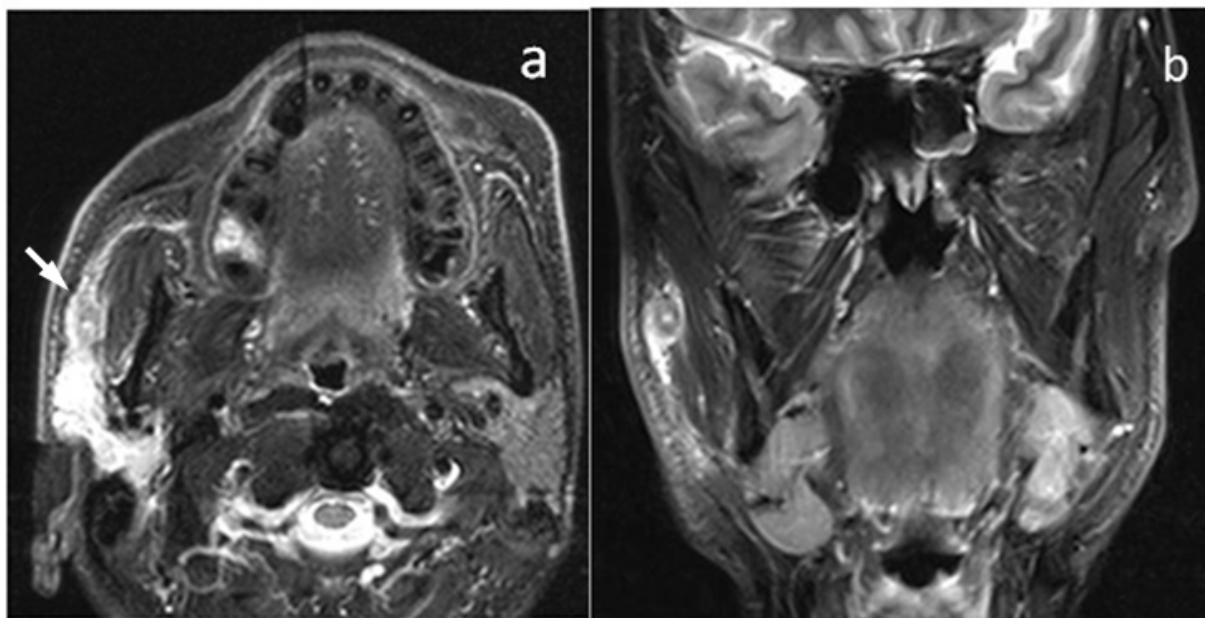


Fig.1

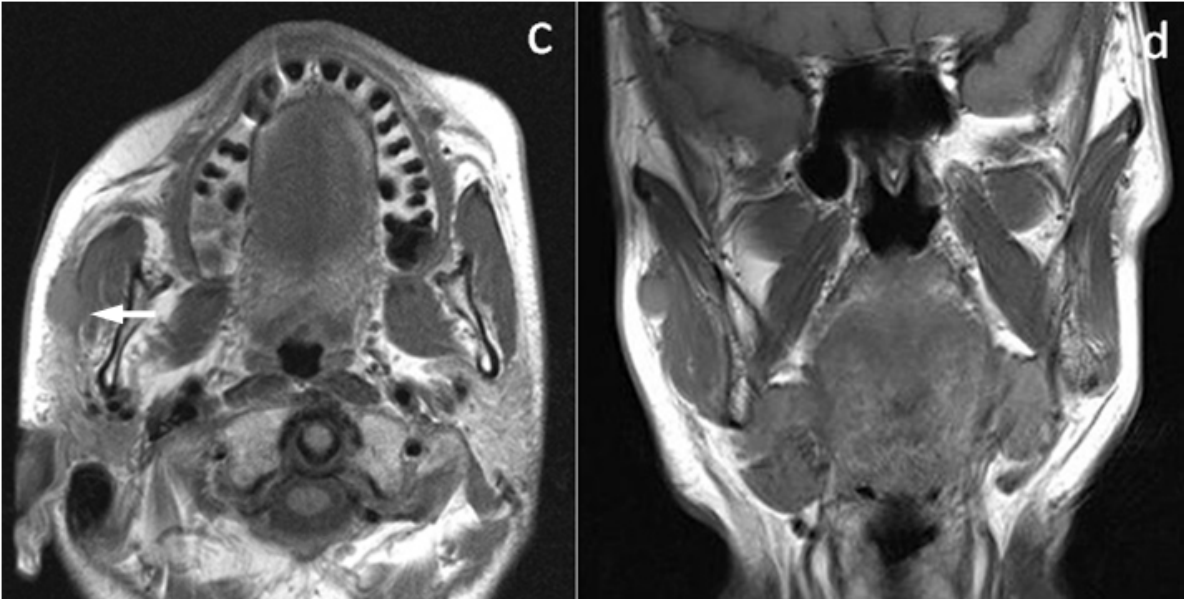


Fig.1

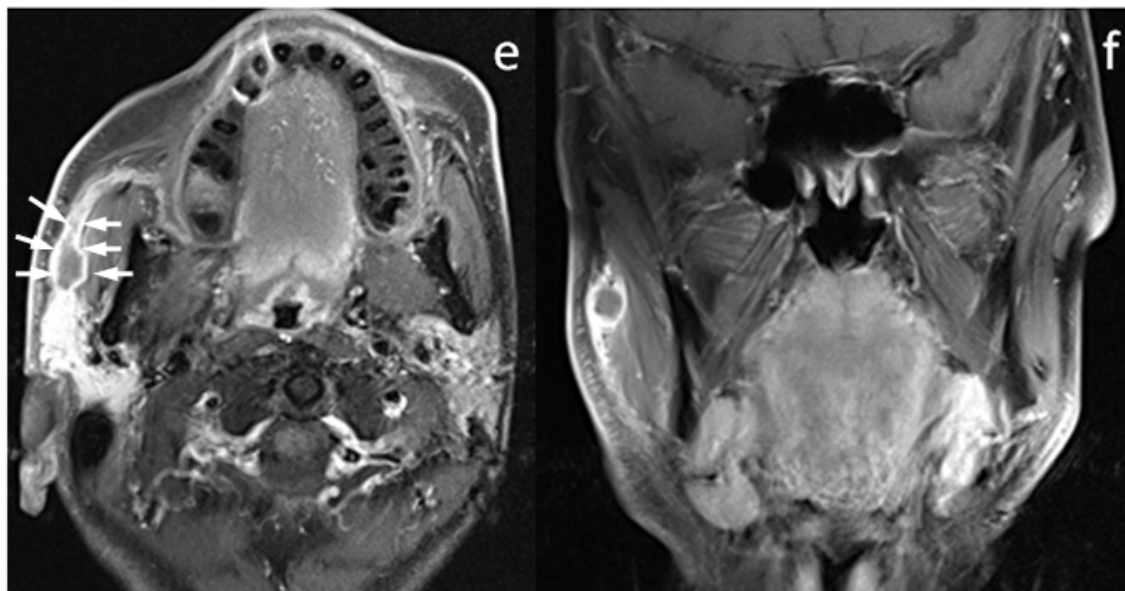


Fig.2

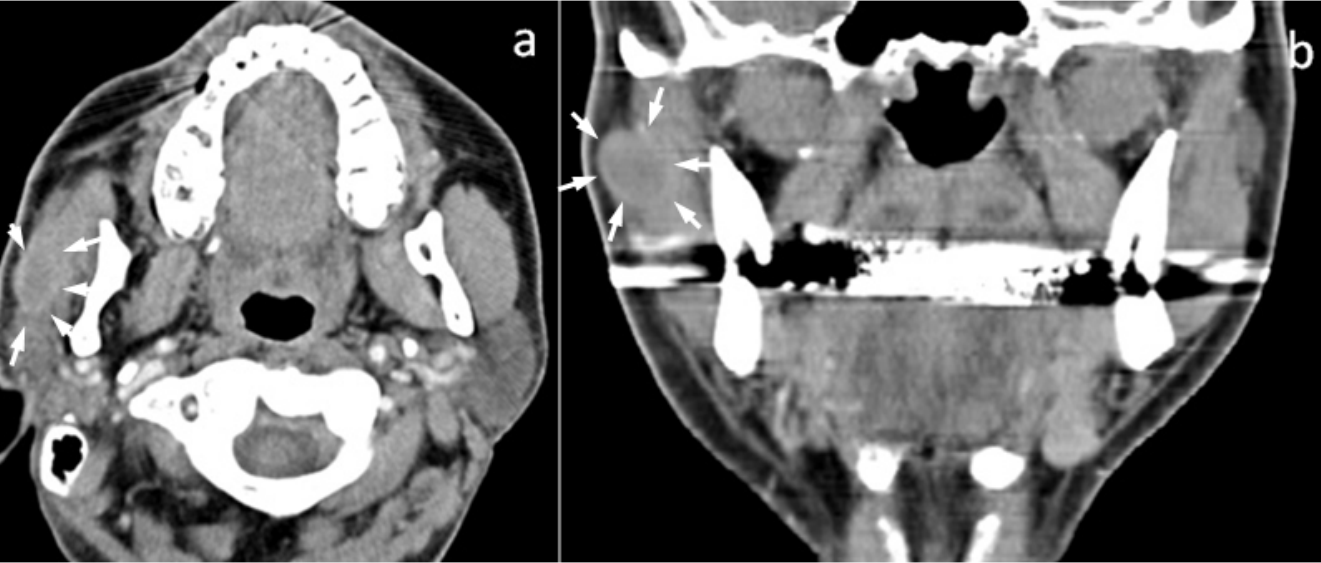


Fig.3

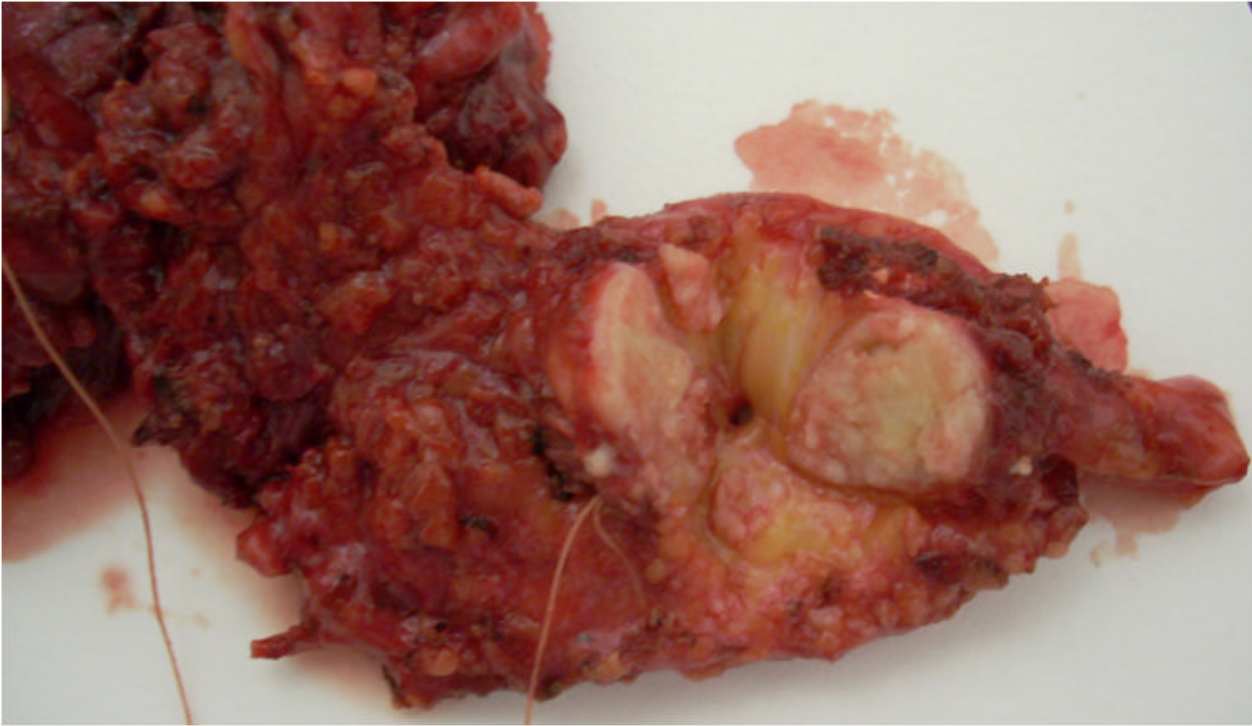


Fig.4

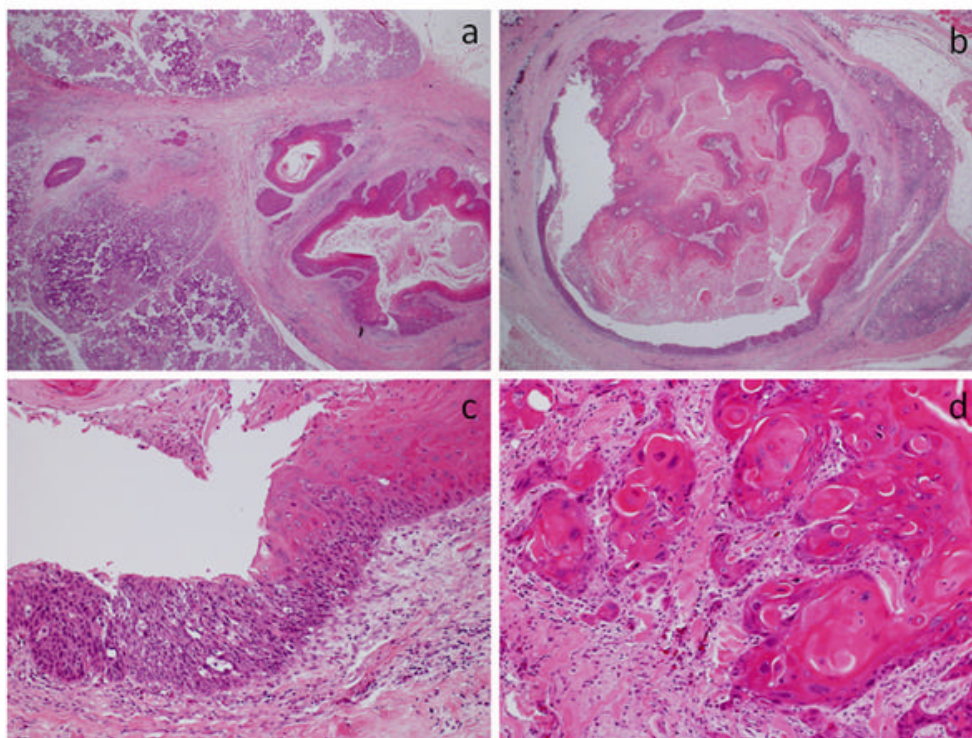


Fig.5

