

Parotid Gland Cancer With First Bite Syndrome Detected via CT-Guided Fine Needle Aspiration Cytology

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Abstract. *Background/Aim:* First bite syndrome (FBS) is a symptom of severe pain at the beginning of a meal that lessens as the meal progresses. It is a common postoperative complication of parapharyngeal space tumors and is rarely reported as the first symptom of parotid carcinoma. The parapharyngeal space is considered a difficult area for approach; hence, preoperative histopathology is often challenging. However, there are hardly any reports on the approach of performing biopsies under computerized tomography (CT) guidance. *Case Report:* A 28-year-old woman presented to our hospital with the chief complaint of pain in the left parotid region since the past year. Contrast-enhanced magnetic resonance imaging of the parotid gland revealed a 10-mm high-signal area on T2-weighted images extending from the deep lobe of the left parotid gland to the parapharyngeal space, which could not be visualized on ultrasound. She was suspected to have a malignant tumor because of the presence of a parotid tumor with FBS. Therefore, she underwent CT-guided fine-needle aspiration cytology (FNAC) and was diagnosed with adenoid cystic carcinoma. The patient underwent left parotid tumor resection and left cervical dissection, and her pain during feeding improved postoperatively. *Conclusion:* In a patient with parotid tumor extending into the parapharyngeal space with FBS as the initial symptom, CT-guided FNAC was

successfully used to diagnose parotid carcinoma. Symptoms of pain, including FBS, should be considered in cases of malignancy. CT-guided FNAC is effective for lesions that cannot be visualized by ultrasound, such as those in the parapharyngeal space.

First bite syndrome (FBS) is a symptom of severe pain at the beginning of a meal that becomes less severe as the meal progresses. It often occurs as a postoperative complication of parapharyngeal space tumors, with an incidence of 9.6% (1). Those without a history of surgery are termed idiopathic FBS but are extremely rare. It is rarely reported as a first manifestation of parotid carcinoma. Most idiopathic FBS cases are caused by tumors. Surgical resection of benign or malignant tumors is an effective treatment option. Postoperative radiation therapy and chemotherapy are often required to treat malignant tumors.

The parapharyngeal space is considered difficult to access because it is an inverted conical space bounded by the skull base above, lateral wall of the pharynx inside, deep lobe of the parotid gland outside, medial pterygoid muscle in front, prevertebral muscle behind, and hyoid bone below (2, 3). Because they are located deep in the face, some areas were not delineated using ultrasound. Parapharyngeal space tumors are relatively rare, accounting for 0.5% of all head and neck tumors (4). Malignant tumors account for 13-26% of cases, with benign tumors accounting for a higher percentage. As the parapharyngeal space is difficult to approach, preoperative fine-needle aspiration cytology (FNAC) often fails to yield adequate results. Therefore, preoperative histopathology is often difficult; however, computed tomography (CT)-guided biopsies have been reported a few times.

Parotid tumors with painful symptoms, including FBS, may be malignant lesions, and preoperative FNAC is recommended. This is a case report of a parotid tumor extending into the parapharyngeal space with FBS that could not be visualized by ultrasound but was diagnosed as a parotid carcinoma by CT-guided FNAC.

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Key Words: CT-guided fine needle aspiration cytology, parotid gland cancer, first bite syndrome, parapharyngeal space.



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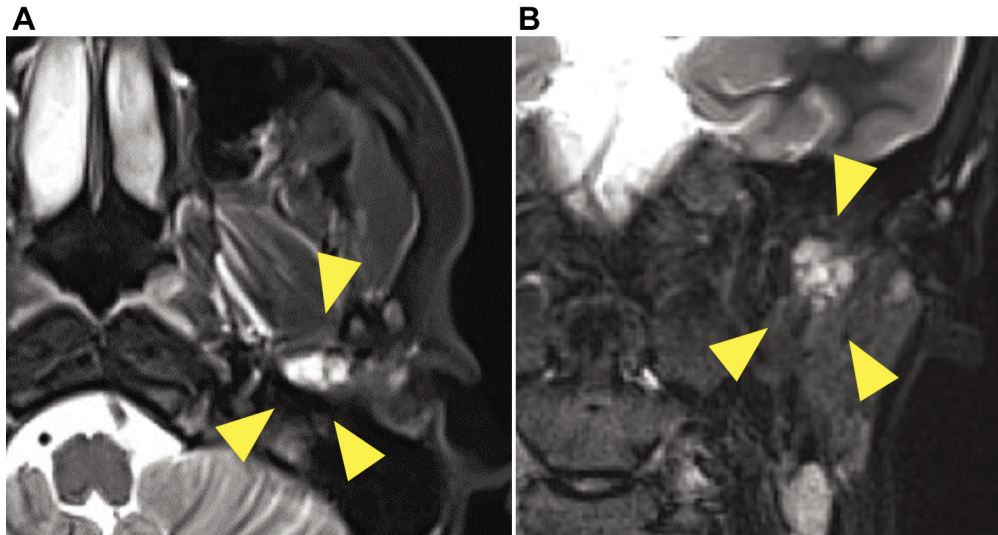


Figure 1. Contrast-enhanced magnetic resonance imaging of the parotid gland. (A) Axial; (B): coronal. A high-signal area with a maximum diameter of 10 mm on T2-weighted images is observed in the deep lobe of the left parotid gland, with a contrast effect at the margins (yellow arrow). The tumor extends into the parapharyngeal space.

Case Report

A 28-year-old woman presented to our clinic in 2021 with left parotid region pain while eating since 2020; the pain had worsened and became uncomfortable since 2019. On initial examination, no obvious mass was palpable in the left parotid region and no facial nerve palsy was observed. No abnormalities were observed in the oral cavity, pharynx, or larynx. Magnetic resonance imaging (MRI) revealed a high-signal area with a maximum diameter of 10 mm on T2-weighted images and a marginal contrast effect in the deep lobe of the left parotid gland (Figure 1A and B). The tumor extended into the parapharyngeal space and was located on the posterior surface of the mandible. No abnormalities were observed in other areas. Because the tumor was located on the back surface of the mandible, it could not be visualized using ultrasound. Fluorodeoxyglucose (FDG)-positioned emission tomography showed no FDG accumulation in the left parotid gland or other areas.

CT-guided puncture aspiration cytology was performed because the tumor could not be delineated using ultrasonography. Contrast-enhanced MRI confirmed the location of the shallow temporal artery, posterior mandibular vein, and facial nerve trunk, and the puncture route was set anterior to the foramen magnum and posterior to the posterior mandibular vein. With the mandibular head as the Merkmal, a 20-G Thermo catheter needle was inserted obliquely upward at the lower part of the mandibular head and lower part of the external auditory canal (Figure 2A-C). As the puncture needle was advanced while obtaining CT

images in real time, the patient felt punctured and complained of pain when the needle reached the tumor. Cytology revealed that the number of specimens required for diagnosis was sufficient. Papanicolaou staining revealed that the cell aggregates had a cribriform structure and mucinous balls; adenoid cystic carcinoma was diagnosed (Figure 3).

In 2021, the patient underwent excision of a malignant tumor in the left parotid gland and left cervical dissection. No invasion of the surrounding tissues or nerve invasion was observed. Fatty tissue in the parapharyngeal space was also resected. Postoperative histopathological examination revealed a cribriform pattern of mild vascular and perineural invasion (Figure 4). A diagnosis of adenoid cystic carcinoma (cribriform) was made. The patient started eating the day after the surgery, and the preoperative pain in the left parotid region improved.

Discussion

We encountered a case in which CT-guided FNAC was successfully used to diagnose parotid carcinoma in a parotid tumor with FBS as the initial symptom that had extended into the parapharyngeal space and was not ultrasound-detectable. CT-guided FNAC was effective in treating lesions that could not be visualized by ultrasound, such as those in the parapharyngeal space.

The salivary glands are dually innervated by sympathetic and parasympathetic nerves. FBS is thought to be caused by sympathetic denervation of the parotid gland, resulting in pain due to overexcitation of myoepithelial cells around the parotid

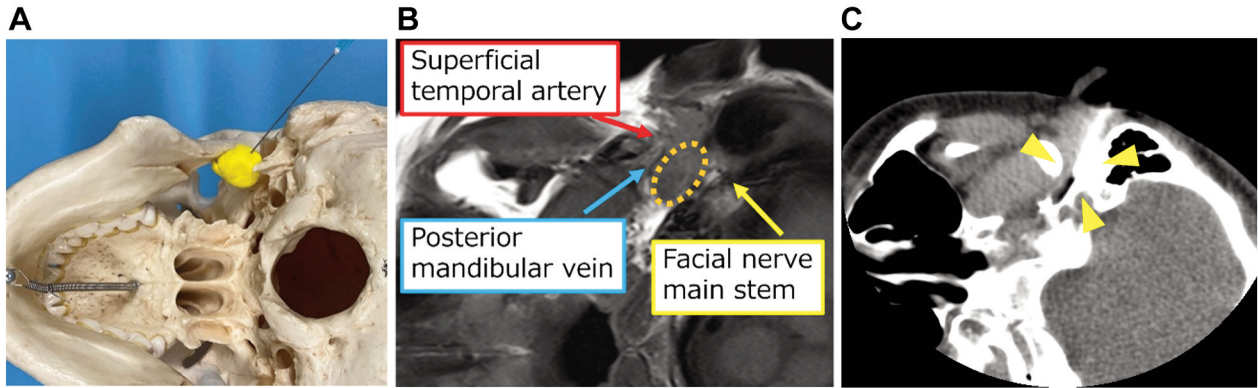


Figure 2. CT-guided fine needle aspiration cytology. (A) The location of the tumor and bone was confirmed using a commercially available skull model. In this model, the position of the tumor was represented with clay, providing a simplified but effective depiction of the tumor's precise anatomical relationship. (B) Contrast-enhanced MRI confirms the location of the superficial temporal artery, posterior mandibular vein, and the main stem of the facial nerve. (C) The puncture route is anterior to the foramen magnum and posterior to the posterior mandibular vein. The mandibular head is used as the meridian, and the puncture is obliquely upward at a level below the mandibular head and below the external auditory canal. The puncture needle is advanced to reach the tumor while obtaining CT images in real-time. CT: Computed tomography. MRI: Magnetic resonance imaging.

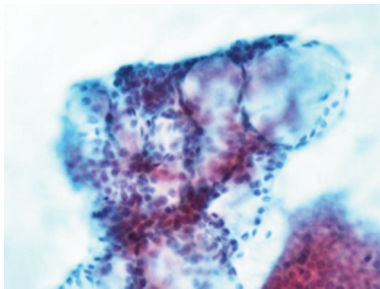


Figure 3. Cytology (Papanicolaou staining). The cellular mass shows a cribriform structure and mucinous balls, and adenoid cystic carcinoma was diagnosed.

gland adeno-hypophysis in response to release of parasympathetic transmitters after oral intake of food (5, 6). FBS occurs as a postoperative complication of parotid deep lobe tumors or parapharyngeal space tumors in >95% of cases; seven cases of parotid tumors with initial presentation of FBS have been reported (7-13). Of these, six were malignant tumors: four in the parotid gland (mucoepidermoid carcinoma, squamous cell carcinoma, adenoid cystic carcinoma, and nonspecific adenocarcinoma), one in the submandibular gland (adenoid cystic carcinoma), and one in the parapharyngeal space (monophasic synovial sarcoma). Surgery was performed in only one patient, and surgery and radiotherapy were performed in five patients. In all cases, FBS disappeared after treatment.

In this patient, postoperative histopathology revealed perineural invasion. Although the cause is unknown, it is possible that the tumor was compressing the sympathetic nerve and that removal of the tumor relieved the compression. To rule

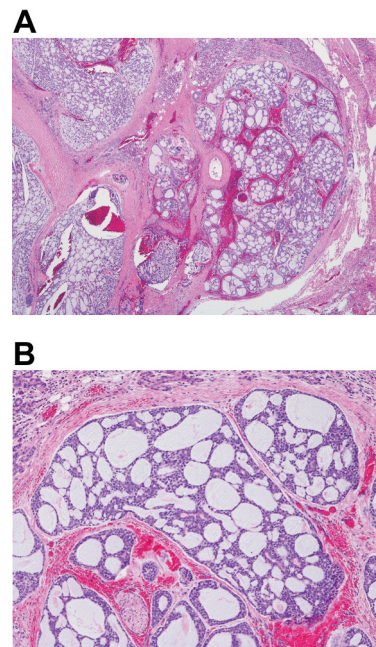


Figure 4. Postoperative histopathology (Hematoxylin and eosin staining). A cribriform pattern is observed, with mild vascular and perineural invasion. A diagnosis of adenoid cystic carcinoma (cribriform type) is made. (A) Magnification 20x; (B) magnification 100x.

out the possibility of malignancy, preoperative histopathology is recommended for salivary gland tumors when the initial symptom is FBS.

Approaches to biopsy parapharyngeal space tumors include transoral, ultrasound-guided, CT-guided, and navigation

techniques. The transoral approach is a simple technique but is limited to the direct visualization of pharyngeal swelling. The positive diagnosis rate is 78-86%; however, the false-negative rate is relatively high, at 19% (14, 15). Ultrasound guidance is often used in head and neck surgery owing to its simplicity and real-time visualization of the surrounding vessels. The accuracy rate is 69-75%. Its disadvantage is the inability to delineate bone and deep crevices (16, 17). CT-guidance can be used even for bony and deep lesions, with a positive diagnostic rate of 66-88% (18). However, prior contrast examination is required to understand the surrounding structures, and the cooperation of a radiologist familiar with head and neck imaging is essential. Navigation-based biopsies have been reported; however, because they must be performed under general anesthesia and the number of cases is small, there are no reports of positive diagnostic rates (19).

CT-guided biopsy is a commonly used technique for the diagnosis of lung tumors in other areas. It can be performed for tumors at almost any site on the trunk and extremities; however, there are few reports of its use in the head and neck region. Complications of CT-guided FNAC include dissemination, pneumothorax, nerve injury, and large vessel injury, with a frequency of 0.003-0.031% (20-22). These correlate with needle size; with needles thinner than 18 gauge, the risk is close to zero. Complications due to vascular injury have been reported, including bleeding from the oral cavity 3 months after biopsy of the masticatory muscle gap, pseudoaneurysm formation in the maxillary artery (22), and rupture of a pseudoaneurysm in the internal carotid artery (23). Vascular complications are considered to be of higher risk in patients after neck surgery or radiotherapy, and caution is required. CT-guided FNAC is highly useful for salivary gland tumors in which malignancy cannot be ruled out, as in the present case, where the lesion cannot be visualized using ultrasound. We hope that this report will be useful for the treatment of salivary gland cancers for which preoperative histological diagnosis is difficult.

Conclusion

We encountered a case of parotid tumor with FBS as the initial symptom that extended into the parapharyngeal space diagnosed as parotid carcinoma using preoperative CT-guided FNAC. Symptoms of pain, including FBS, should be considered as possible malignancies. CT-guided FNAC is effective for lesions that cannot be visualized by ultrasound, such as those in the parapharyngeal space.

Conflicts of Interest

The Authors declare no conflicts of interest, potential conflicts, or financial relationships.

Authors' Contributions

IO and MH conducted case studies. MH wrote the manuscript and prepared the figures. IO, MH, AS, SA, TY, DY, and KT oversaw patient treatment. IO, MH, AS, SA, and TY were involved in data collection. All Authors discussed the results of the case report, commented on the article, and approved the final version for publication.

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