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Case Report

Parapharyngeal teratoma in an adult: a case report and literature review

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Abstract: We report an unusual case of a 61-year-old Chinese man who presented with a 15 years of foreign body sensation and unclear speech, and 1 year of dysphagia. Radiographic imaging revealed a large space occupying lesion of mixed fat density in the left parapharyngeal space, consistent with a teratoma. The mass was completely resected with an intact capsule and characterized by a soft, irregular shape, and local bone-like stiffness. The diagnosis of teratoma was confirmed by pathological examination. Microscopic examination showed a mesoderm component, containing diffuse mature adipose tissue with distribution of sparse fibrous bands and mature bone tissue with mature cortical bone and marrow. During 1 year of follow-up, the symptoms of pharyngeal foreign body sensation and difficulty swallowing disappeared, with no hoarseness or aural fullness.

Keywords: Teratoma, parapharyngeal space, CT scan, pathological examination, surgical resection

Introduction

The incidence of teratoma is about one per 4,000 live births each year, only 3%-10% occur in the head and neck region [1-4]. It most commonly appears in the neck followed by nasopharynx. It can be divided into three categories: mature teratoma, immature teratoma, and malignant teratoma. The clinical manifesttions of benign teratoma are usually foreign body sensation and affected pronunciation. Head and neck CT scan can help to locate and diagnosis the tumor, that is, a lesion can be highly suspected of being a teratoma when the tumor presents with a clear boundary and mixed signal involving both fat and calcification. The therapeutic principle for a benign teratoma in the parapharyngeal space is complete resection. This study reports a case of a mature teratoma in the parapharyngeal space treated in our department, with a review of the anatomical location, pathology characteristics, clinical manifestation, imaging examination, and treatment principles of parapharyngeal space teratoma.

Case report

A 61-year-old male patient was admitted to our hospital complaining of a feeling of swallowed foreign body sensation and unclear speech for 15 years as well as difficulty swallowing for 1 year. A parapharyngeal mass was found on the left side. He had been treated by swelling puncture and incision 15 years previously, and about 200 ml of bloody fluid had been removed. The subjective symptoms were relieved, and the patient did not receive further treatment. One year previously, the self-reported feeling of a pharyngeal foreign body was aggravated again, accompanied by difficulty swallowing and mild left aural fullness. The patient had no fever, headache, sore throat, dyspnea, laryngeal spasm, or hoarseness. He also had no history of hypertension, diabetes, or heart disease.

Physical examination revealed that the soft palate and lateral wall of the pharynx on the left side were upheaved obviously close to the midline, and the uvula was slightly tilted to the right. The mass extended from the top of the nasopharynx to the lower level of the tonsil and was soft with no tenderness. The left eardrum was compressed, but not perforated. The right eardrum was normal. The results of a bilateral Rinne test were: air conduction > bone conduction; those on the Weber test were: left; pure tone listening examination showed mildly conductive deafness in the left ear. Enhanced com-

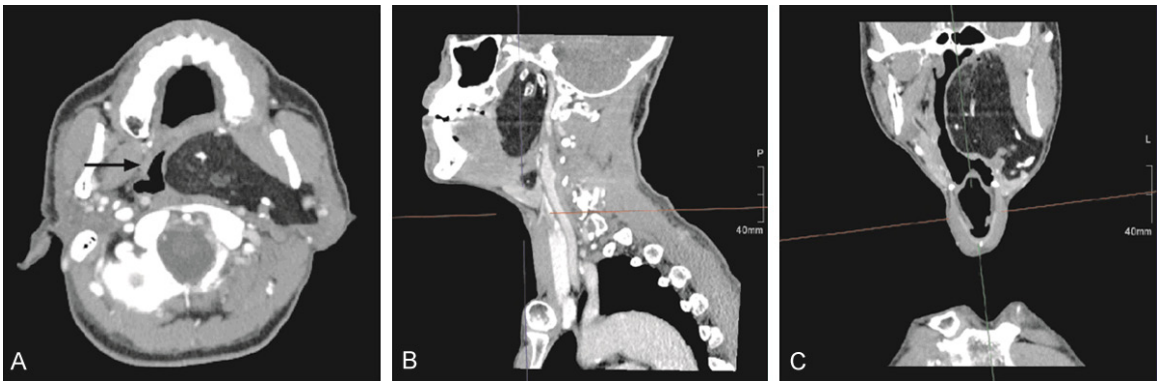

Figure 1. CT scans of the lesion before surgery. Axial (A), sagittal (B) and coronal (C) demonstrated a large space occupying lesion (arrow) with mixed fat density in the left pharyngeal space, considered a teratoma.

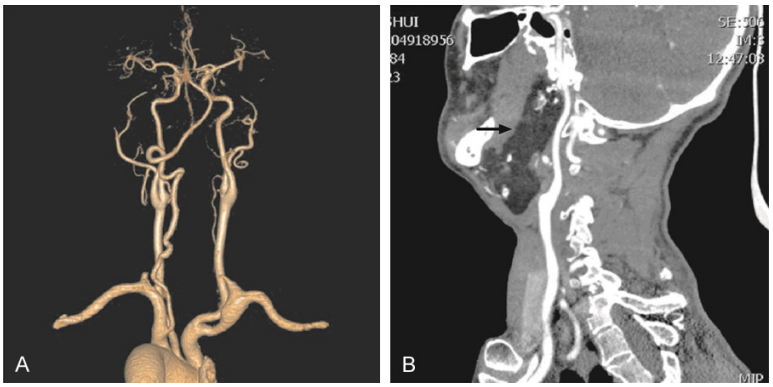

Figure 2. The relationship of the tumor and the artery. CT scans showing a mixed density, space occupying lesion (arrow) located on the left side of the pharynx, pushing the left internal and external carotids to the left rear.

puted tomography (CT) of the neck revealed a large space occupying lesion of mixed fat density in the left pharyngeal space, consistent with a teratoma (**Figure 1**). The cervical vascularized enhanced CT (CTA) indicated a mixed density occupying lesion on the left side of the pharynx, pushing the left internal and external carotids to the left rear (**Figure 2**).

After complete preoperative examination, the patient underwent resection of the left pharyngeal space neoplasm under general anesthesia from the external cervical approach (**Figure 3A**). The mass envelope was complete. The tumor was separated along the envelope carefully, and no adhesion was found between the mass and internal jugular artery and vein. After cutting off part of the styloid process, the mass was completely removed with an intact capsule and characterized by a soft, irregular shape, and local bone-like stiffness (**Figure 3B**). After suturing the cervical incision, a small amount of blood clot was observed in the mouth. After the blood clot was absorbed, a small break about 1 cm long on the lateral side of the left palatal tongue arch was sutured. During follow-up, the symptoms of pharyngeal foreign body sensation and difficulty swallowing disappeared, with no hoarseness or aural fullness.

Microscopic observation showed a mesoderm component, containing diffuse mature adipose tissue with distribution of sparse fibrous bands and mature bone tissue with mature cortical bone and marrow (**Figure 4A**). The entoderm component had complete salivary glands, including mucinous gland vesicles, serous gland vesicles, mixed adenoids, and ducts (**Figure 4B**). The diagnosis of mature teratoma was made.

Discussion

The paralopharyngeal space is a potential gap presenting as an inverted cone-shape between the medial pterygoid, the deep parotid gland, and lateral pharyngeal wall. The parapharyngeal space extends from the temporal and sphenoid bones of the skull base down to the hyoid bone. Its medial boundary is the upper pharyngeal constrictors, and its lateral boundary is the mandibular ramus. It is located after the cervical vertebrae and before the pterygo-

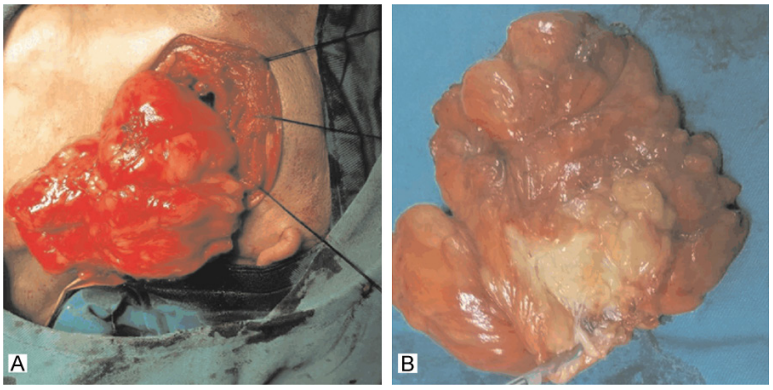

Figure 3. Imaging of the tumor. Intraoperative image of the tumor with intact membrane (A). The mass was completely removed with an intact capsule and characterized by a soft, irregular shape, and local bone-like stiffness (B).

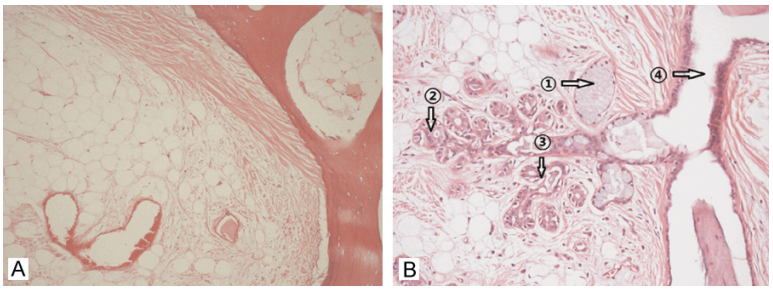

Figure 4. H&E staining of the tumor slice. (hematoxylin and eosin staining ×100) Mesoderm component containing mature adipose tissue diffused distributing sparse fibrous bands and mature bone tissue with mature cortical bone and marrow (A). (hematoxylin and eosin staining ×200) Ectoderm component showing complete ① mucinous gland vesicles, ① serous gland vesicles, ③ mixed adenoids and ④ ducts (B).

mandibular raphe. It is divided into the prestyloid and poststyloid spaces bound by the styloid process and its adhesion structures, such as the stylohyoid muscle and stylopharyngeal muscle. A tumor occurring in parapharyngeal space is rare, accounting for about 0.5% of head and neck tumors [5]. The location of a tumor in different compartments of this space usually indicates the potential histologic etiologies of the lesion. A tumor in the prestyloid space is often derived from a salivary gland, such as the parotid pleomorphic adenoma [6]. Tumors in the poststyloid space are mostly paraganglioma, schwannoma, and other rare malignant lesions.

Teratomas originate from totipotential stem cells [7]. According to their histological features, they can be divided into three categories: mature teratoma, immature teratoma, and malignant teratoma. Immature and malignant teratomas are composed of undifferentiated tissue and usually have a high potential for malignancy [8]. Mature teratomas are also referred to as dermoid cysts, which usually have a smooth and intact envelope. Teratoma is a true neoplasm that may include tissues from all three blastoderms and develop independently of the host. Histopathologically, the capsule wall contains squamous epithelium and its appendages, such as hair follicles, sebaceous glands, and sweat glands [9, 10]. In addition, capsule contents contain mature nerve tissue derived from ectoderm, such as glial tissue, as well as fat, cartilage, and smooth muscle tissues from mesoderm and ectoderm. They are considered to grow from misplaced embryonic, pluripotent germ cells that lose impact during embryologic development [4].

Most teratomas reported in the literature occurred in the sacrococcygeal region, gonad, and other sites such as the mediastinum and retroperitoneum. Most teratomas occurring in the pediatric populations are found within the pelvis and are typically benign in nature. Head and neck teratomas occurring in newborns and infants mainly causes respiratory distress [11], and those in adults may present as asymptomatic throat and neck neoplasms. Cases of huge and benign tumors are usually associated with sleep apnea and difficulty breathing and eating. To date, fewer than 40 cases of head and neck teratomas in adults have been reported in the literature. In the present case, the diagnosis and treatment of the patient was a process lasting more than 10 years, and the tumor caused obvious pharynx foreign body sensation and affected his pronunciation, which conformed to the clinical manifestations of a benign teratoma.

Head and neck CT examination can help to locate a tumor in the pharynx. In addition, the

CT manifestation of a teratoma has important diagnostic value; that is, a lesion can be highly suspected of being a teratoma when the tumor presents with a clear boundary and mixed signal involving both fat and calcification [4]. However, lipoma and liposarcoma with radiologic evidence of fat lesions should be carefully distinguished. Intravenous contrast is also commonly used together with CT to help demonstrate the displacement of the carotid artery. A teratoma typically presents local low attenuation representing fat, foci of high attenuating calcifications, and diffuse heterogeneous areas with soft tissue densities. Moreover, since magnetic resonance imaging (MRI) provides better soft tissue resolution in the head and neck, it has more advantages for clarifying the surgical resection range compared with CT.

Conclusions

The therapeutic principle for a benign teratoma in the pharyngeal space is complete resection. An intraoral or transmandibular approach can be selected according to the infiltration of the tumor site and the surrounding tissues involved. The parapharyngeal space tumor specimen should not be taken out through the mouth to avoid damage to blood vessels and nerves. In addition, it may destroy the integrity of the membrane, leading to the dissemination of tumor cells. This patient had received intraoral incision drainage several years previously, which was risky. Moreover, the local integrity was damaged, resulting in mucosal damage in the scar adhesion during resection. The transcervical approach has the advantages of full exposure and reducing blood vessel and nerve damage. Therefore, it is frequently used for the detection, diagnosis, and treatment of benign tumors in the parapharyngeal space. The range of resection should be expanded when the tumor presents malignant potential. Once diagnosed with an immature or a malignant teratoma, patients commonly need postoperative adjuvant radiotherapy and chemotherapy. Our patient showed no evidence of recurrence after 1 year of follow-up.

Disclosure of conflict of interest

None.

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