Case Report

An organized hematoma in the right parapharyngeal space: a case report and literature review

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Abstract: Organized hematoma is a relatively common condition that can occur in various locations throughout the body. There have been sporadic reports of organized hematomas in the literature, reported in locations such as the maxillary sinus, thyroid gland, and subdural cavity, which are common in the head and neck. Studies have shown that the mechanism of organized hematoma formation may be related to trauma, surgery, hemorrhage, vascular disease, or hematological disease. The composition of organized hematomas is mostly identical, the center is formed by a large number of organized blood clots which are surrounded by granulation tissue and dense fibrous connective tissue. Parapharyngeal space (PPS) is a latent fascial space deep in the lateral superior neck of the pharynx. Pathological changes in the PPS are rare, and most of them are benign tumors. An organized hematoma in the PPS is extremely rare. Only one case has previously been reported. Here, we report a second case in which a fine needle aspiration biopsy indicated a benign adenoma in the right PPS. The findings help to expand the differential diagnosis of PPS tumors and deepen the understanding of PPS organized hematoma, a rare benign lesion.

Keywords: Organized hematoma, parapharyngeal space, tumor

Introduction

The incidence of parapharyngeal space (PPS) tumors is low. Worldwide, only 0.5% of head and neck tumors originate in the PPS, 80% of which are benign [1]. Of the primary tumors of PPS, 50% originate in the salivary glands and represent mostly pleomorphic adenomas from the deep lobe of the oblongata, 30% are neurogenic tumors, typically from IX-XII schwannoma of the brain and sympathetic nerves, and 20% originate in soft tissue [2]; they include angiomomas, lipomas, teratomas, rhabdomyomas, and fibromas. An organized hematoma occurring in the PPS is extremely rare, and only one case has been reported thus far [3]. This type of tumor is difficult to detect in the early clinical stage because of its unique anatomical location [4]. The determination of the specific mechanism of PPS hematoma in the case reported in 2009 was not clear. The patient denied trauma, hemorrhage and blood system disease, the medical history and preoperative laboratory examinations did not indicate any definitive abnormality. The final diagnosis of organized hematoma was based on pathological examination. The specimen consisted of a central blood clot, inflammatory exudates, necrotic region, calcified tissue, granulation tissue, and external fibrous connective tissue [5].

Case report

A 54-year-old male presented with a slurred speech that lasted for two months, without any history of blunt trauma, hemorrhage, or hematologic disease. Physical examination showed a huge bulging tumor with smooth surface located in the right PPS area. The lesion appeared soft, measuring about 3 × 2.5 × 3 cm, the epiglottis was without congestion and swelling, the mobility, double vocal cord appearance and the motion was normal. Laboratory data, including blood count, biochemistry, electrolyte levels, platelet count, and coagulation time were normal. Computerized tomography (CT) scan demonstrated an oval mass on the right side of the pharynx and palate, 5.0 × 6.5 cm in size, with a poorly defined margin, extending from the lower part of the caput mandibular
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down to the epiglottis and the right side of the PPS where the right pyriform fossa becomes shallow (Figure 1). The enhanced CT scan showed an oval mass of about 5.0 × 3.5 × 6.5 cm in size on the right side of the neck, with a slight uneven enhancement and a clear margin. The mass reached the level of the foramen magnum of the occipital bone and descended to the epiglottis. The right pharyngeal recess and the pharyngeal orifice of the eustachian tube were shallow, the right PPS, the right piriform fossa, nasopharynx, and oropharyngeal cavity were narrowed (Figure 2). Magnetic resonance imaging (MRI) showed an oval mass of about 7.0 × 5.5 × 4.0 cm on the right side of the oropharynx. The lesions produced slightly abnormal signals, while the STIR phase produced mostly mixed and high signals. The lesion had a regular shape, smooth edges, and low signal interval shadows were visible in the lesion. The mouth floor, oropharynx, and nasopharynx were compressed and partially occluded (Figure 3). Fine needle aspiration cytology (FNAC) was performed before the surgery and showed a large number of red blood cells and a small number of adenomatous cells. The entire mass and its fiber wrap were completely removed via transcervical and transoral
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approach under general anesthesia with tracheotomy and intubation. During the procedure the right pharyngeal wall and soft palate appeared to be bulged, the lesion spanned from the nasopharyngeal isthmus down to the upper epiglottis, and the tumor was located in the right PPS. The lesion measured about 6 × 5 × 3 cm in size, had a smooth surface, medium texture, and was dark red. The surrounding tissue border was clear, with a gray-white and brown section, and the right submandibular gland was enlarged, and the submandibular triangle had two enlarged lymph nodes (Figure 4). Postoperative pathological examination showed that the tissues contained fibrous connective tissue, granulation tissue, and organized blood clots, and the lesion corresponded to an organized hematoma (Figure 5). The patient recovered well after the operation, and there was no recurrence after a half year follow-up.

Discussion

The etiology of organized hematoma in the PPS remains unknown. Hsu first reported this disease in 2009 but did not elucidate the specific cause and mechanism of the formation of organized hematoma. The patient denied any history of trauma, hemorrhage, or hematological diseases, and the results of preoperative laboratory tests, including blood coagulation and platelet count, were within the normal ranges [3]. Organized hematomas have been reported to occur in multiple tissues throughout the body, with those in the head and neck occurring typically in the maxillary sinus [5]. In 2013 a study showed that the clinical features of five cases of maxillary sinus organized hematoma, concluding that the main clinic pathological characteristics of maxillary sinus organized hematoma can be summarized as a combination of multiple factors, such as mixed hemorrhage, vasodilation, cellulose exudation, fibrosis, hyaline degeneration, and capillary hyperplasia. The expression of periosteum, CD31, and CD34 in the hematoma was also observed by immunohistochemistry [6]. In addition, we have retrospectively analyzed the cases of other organized hematomas and found that most of the causes of these lesions were related to trauma, hemorrhage, angiogenesis, and abnormalities of coagulation [7, 8]. Other investigators studying hematoma of the cranial bone...
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block concluded that the mechanism of formation of organized hematoma is due to the lack of decomposition of hemosiderin before liquefaction of the hematoma. The absence of this process leads to an increase in the fibrous substance and, therefore, the inability of the hematoma to become completely liquefied [9]. As a result, a capsule is formed, providing the structure for hematoma organization and localized mass. It is also hypothesized that the formation mechanism of epidural hematoma in the elderly is related to brain atrophy, enlargement of subarachnoid space, and a compensatory increase in cranial space [10]. Ralph reviewed seven cases of intradiploic hematoma of the skull and found that all patients had a history of mild head trauma accompanied by a slow expansion of the skull [11]. Therefore, it can be speculated that the disease may be caused by abnormalities of blood vessels or idiopathic bleeding. After a small amount of bleeding occurs, the blood coagulates to form a hematoma, in which a fibrous fascicular membrane is formed approximately two weeks later. The fibrous fascicular membrane contains a large number of new capillary networks, and the endothelial cells of the new vessels possess large intercellular spaces and abnormal permeability. At the same time, local eosinophil degranulation activate fibrin kinase, promoting fibrinolysis inhibiting platelet aggregation and result in a gradual increase in the hematoma size. Later, the capsule of the hematoma undergoes vascular embolism necrosis, connective tissue degeneration and calcification, and the intracapsular hematoma becomes gradually organized, forming a large PPS hematoma. However, more evidence is needed to elucidate the specific causes of PPS organized hematoma.

Diagnosis of the early stages of organized hematoma in PPS is challenging due to the unique anatomical location. It has been documented that the tumors often grow to at least 2.5-3 cm before they are detected, and the main clinical symptom of tumors of the PPS is a painless mass in the neck or the oropharynx.

It has been reported that 20% of PPS tumors are found incidentally, and 15% of benign PPS tumors may be associated with preoperative cranial nerve deficits [12]. Medical history and physical fitness provide reliable evidence for this rare case, but the location of PPS is deep, and the scope of detection is limited by the unaided eye. It is impossible to observe the actual size of the tumor, the extent of invasion, and its relationship with surrounding tissues, like muscles, nerves and blood vessels. Therefore, the use of imaging techniques is critical for the diagnosis and preoperative evaluation of parapharyngeal space tumors.

MRI provides higher-resolution images of soft tissues and can more clearly determine the origin and extent of the tumor, providing a reference for the preliminary determination of the source of the tumor. On the other hand, enhanced CT has a higher resolution for the distribution of blood vessels within and around

Figure 5. Photomicrographs of the sections of the tumor showing a cavity filled with fibrin, blood cells (blue arrows), granulation tissue (white arrows), and fibrous connective tissue (black arrows). H&E staining. Original magnification: (A) 40 ×; (B) 100 ×.
the tumor, can show the relationship between the tumor and the cervical vessels or other nerves more clearly, and can visualize whether the tumor has eroded the bone. According to the report of Lee, the typical manifestation of organized hematoma detected by enhancement CT is a heterogeneous or homogeneous calcification [13]. It is also considered that the CT findings of rare chronic organized subdural hematoma present in the head can be manifested as mixed and low-density occupancy [8]. On the other hand, MRI examination also provides certain specificity in the imaging diagnosis of organized hematoma. Kim demonstrated the existence of heterogeneous and high-signal changes and heterogeneous and low signal in the center of the sinuses organized hematoma [14]. In two cases of maxillary sinus hematoma, MRI showed inhomogeneity signal changes from low to high on T2WI and slightly high on T1WI [15]. Imaizumi S pointed out that the intratumoral mixing with cord-like septa is an important feature of the MRI images of head organized hematomas [16]. In our case, MRI scans showed that there were slightly long or isoT1, long or isoT2 abnormal signals, and STIR yielded a mixed signal with high signal intensity. Additionally, in agreement with the work of Imaizumi S, the lesion was regular in shape, and the edges were smooth with low signal intervals in the center. Thus, although the imaging manifestations of organized hematoma can be conclusive, differences in individual performance are present.

The treatment of giant hematoma of PPS is the same as in the case of a benign tumor in this location, and the gold standard of therapy is surgical extirpation. Before complete excision of the tumor, FNAB is recommended. However, this procedure does not always provide an accurate diagnosis, and often complete removal of the tumor is needed to determine the final diagnosis and design the treatment. Interestingly, in the case presented here, the preoperative FNAB indicated a benign adenoma, and only the final complete resection allowed reaching the diagnosis of an organized hematoma. Currently, four approaches are used for the resection of a PPS tumor: the transcervical lateral approach, the neck parotid gland approach, the neck side of the mandibular fissure approach, and the transoral approach [17]. The selection of the approach depends on the size of the tumor, its location, and relation to the major surrounding vessels, the possibility of malignancy, and the experience of the surgeon. In the present case, the CT scan documented an elliptical mass of about 5.0 × 3.5 × 6.5 cm in the right neck, occupying the space from the level of foramen magnum, down to the epiglottis involving the right PPS, pyriform fossa, nasopharynx and oropharyngeal cavity. Considering the difficulty to completely expose the tumor using a transcervical lateral or transoral approach, it was problematic to protect important blood vessels and nerves during operation. Therefore, a transcervical incision and combined intraoral approach were used to remove the tumor. This approach can directly expose the upper and lower levels of the tumor, protect the carotid artery and nerve, and allows an easy extension of the incision to the parotid gland and mandible.

Differential diagnosis of organized hematoma in PPS is difficult because the imaging findings frequently lack specificity. Clinical history of trauma, bleeding, surgery, and hematological disease may be a significant clue for organized hematoma of the PPS. Interestingly, as reported in this case of PPS hematoma, the patient denied any previous trauma, surgery, or bleeding tendency.

In conclusion, organized hematoma in the PPS is rather rare in the clinic, with only two known cases, including the present one, currently reported. Chronic organized hematoma in the PPS is a lesion that must be differentiated from various types of tumors which are located in the PPS, in particular when patients have a history of trauma or coagulopathy. Surgical resection represents the best choice of treatment.

Disclosure of conflict of interest

None.

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