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Surgical management of recurred hibernoma of the neck

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August 17, 2022

Abstract

Cervical hibernoma is a rare, benign lipogenic tumor. Surgical removal of cervical hibernoma is needed when it causes symptoms such as dyspnea or dysphagia due to its mass effect, or when malignancy cannot be excluded. During surgery, en bloc resection is crucial to prevent hemorrhage and recurrence.

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Keywords: hibernoma, lipoma, brown fat, lipogenic tumor, cervical mass

Key Clinical Message: This case review shows a successful removal of recurred hibernoma in neck. En bloc resection is important to prevent recurrence and bleeding.

Introduction

Hibernoma is a rare, benign lipogenic tumor that mostly occurs in thigh. It occurs even more rarely in head and neck area, and there are only few case reports of head and neck hibernoma. This article is a retrospective review of a successful removal of recurred hibernoma in neck.

While managing patients with lipogenic tumor, clinical suspicion should be primarily needed and histological, imaging work-ups should be followed to exclude lipomatous malignancy. Surgical removal of cervical hibernoma is needed when it causes symptoms such as dyspnea or dysphagia due to its mass effect, or when malignancy cannot be excluded. During surgery, en bloc resection is crucial to prevent hemorrhage and recurrence.

Case report

A 16 year old female was incidentally found to have mediastinal widening in plain chest X-ray took at medical check-up and referred to thoracic surgery department of Seoul National University Hospital (SNUH) at 2019-04-10. She denied any underlying diseases or family history of malignancy, and she had never experienced smoking or alcohol. She had dysphagia, weight loss and exertional dyspnea at the time of the first visit, but her symptom was not severe and neglected until plain chest X-ray was taken. Her initial height was 160cm and body weight was 42.5kg.

Chest CT taken at outside hospital showed about 9.5x6.1cm sized fat containing mass lesion without calcification in the lower neck to posterior upper mediastinum, and tracheal compression was observed. The differential diagnosis made by radiologist was well differentiated liposarcoma or immature teratoma. F-18 FDG PET/CT was taken and it showed mild uptake (˜1.9) at aforementioned mass, with metabolic defect in fat portion (Figure 1). The core needle biopsy of the mass was done, and the pathologic report confirmed lipogenic tumor with some brown fat cells, suggestive of hibernoma.

Mediastinal mass excision via full sternotomy under V-V ECMO support was done by thoracic surgery team. Anesthesiologist recommended V-V ECMO support because there was mid tracheal compression more than 50%, with diameter lower than 5mm, through which plain E-tube ID 3.0mm could barely pass. Before the anesthesia induction, fiberoptic, rigid bronchoscope was prepared and high flow nasal cannula was applied for the possibility of emergent airway collapse. Under MAC anesthesia, the V-V ECMO was inserted with 17Fr catheter at right and left femoral vein, then the general anesthesia with endotracheal intubation with plain ID 6.5mm tube was followed. There was no difficulty during the intubation process, and the intubation depth was 22cm. Airway patency was checked with fiberoptic bronschoscope.

During the main procedure, approach through full median sternotomy was made, and large mass through superior aspect of anterior mediastinum to anterior neck was identified. The mass was well circumscribed and lobulated, and the adhesion to nearby tissues was minimal. There was no evidence of vessel, nerve, pleural invasion, and delicate dissection with complete removal was performed. Bilateral recurrent laryngeal nerves were identified and preserved. The patient was transferred to pediatric intensive care unit after the surgery, and was discharged at postoperative day 5 without any complication.

The size of the mediastinal mass was 9.6 x 7.9 x 3.8cm, and the final pathology confirmed mediastinal hibernoma. On gross examination, resected mediastinal lesion contained fatty tissue-like yellowish mass measuring 5.3 cm. The mass was composed of polygonal cells resembling brown fat with multivacuolated cytoplasm, admixed with mature adipocytes. The lesion was also characterized by fibrous septae and myxoid stroma, suggestive of myxoid variant. Small, delicate, branching capillaries were noted. Nuclei were small with no significant atypia, and necrosis was not observed. Immunohistochemistry for CD31 showed staining in hibernoma cells as well as capillary endothelial cells (Figure 2).

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Regular follow up at outpatient clinic was done with chest PA, and there was no evidence of recurrence. Initial symptoms such as dysphagia and dyspnea were also disappeared, and she had no complaint. She also gained weight, and her body weight was 47.4kg at 2022-01-10. However, the chest CT taken at 2020-08-26 showed small attenuating lesion at left supraclavicular area. The size of the mass was increased to about 2.2cm, which was suspicious for residual mass or recurrence of hibernoma. Due to its location, the patient was referred to Otolaryngology department at SNUH.

2022-01-10 neck CT was taken, and the size of the left level VI mass was increased (2.2 -> 2.6cm) (Figure 3). In addition, 1.6cm mass was incidentally identified at right submandibular gland posterior aspect, and followed ultrasound gun biopsy confirmed pleomorphic adenoma.

Eventually, left level VI neck dissection with right submandibular gland mass excision was done at 2022-01-25. Electromyography tube was intubated for intraoperative recurrent laryngeal nerve monitoring. About 5cm midline horizontal incision was made along the skin crease at the level of thyroid gland. During the removal process, the mass was well circumscribed with capsule and pinpoint capsule violation was observed, but there was no definite spillage of tumor content. Delicate dissection was performed to save nearby structures (Figure 4). Eventually, complete removal was achieved, and left vagus nerve, carotid vessels, thyroid gland were saved. Left recurrent laryngeal nerve was not identified due to severe adhesion, but the patient's vocal fold movement was intact postoperatively. Suspicious fat tissue nearby main mass was also removed. The right submandibular gland mass was also removed in a routine manner.

The recurred tumor at left level VI was also confirmed as hibernoma, which had similar histologic features with more pronounced myxoid stroma compared to mediastinal mass (Figure 2). Removed fat tissue adjacent to main mass was diagnosed as mature adipose tissue. The right submandibular gland was confirmed as pleomorphic adenoma with clear resection margin.

The patient was discharged at postoperative day 2 without any complications, including vocal fold palsy, hematoma, wound infection, etc. There was no evidence of recurrence at 6 month follow up neck CT.

Discussion

Hibernoma is a rare, benign tumor which consists of cells that histologically resemble brown fat cells. It was first reported by Merkel in 1906 and then named by Grey as "hibernoma" in 1914, because of its histological similarity to the brown fat of hibernating animals [2]. Unlike typical adipose tissue, brown fat has abundant vascularity to generate heat [3]. Brown fat usually disappears after 8 weeks of life, and it is slowly replaced by white fat [4].

Hibernoma accounts for only 1% of all benign lipomatous tumors and are usually asymptomatic unless it compresses nearby structures with bulky mass. It is most commonly identified in the thigh, but it can also occur anywhere with scattered brown fat, such as bone, retroperitoneum, scalp, neck, axilla, shoulder, thorax, breast, stomach, etc. [2]. Its prevalence peaks in the fourth and fifth decades of life, and gender predominance is controversial among articles [3, 4].

Due to its low incidence, it is sometimes misdiagnosed as well-differentiated liposarcomas, or myxoid liposarcomas. On computed tomography (CT) scans, hibernomas sometimes present as well circumscribed, hypodense lesions with linear septations, which indicates high vascularity [3]. On MRI, hibernomas can be isointense or relatively hypointense to the surrounding fat on both T1 and T2, and diffuse heterogenous enhancement can be identified due to hypervascularity of the mass. On F-18 FDG PET/CT scan, hibernomas show uptake because of metabolically active brown fat tissue. However, none of images listed above are pathognomic, and this also hinders the differentiation of hibernoma with sarcomas [2].

Therefore, the pathologic diagnosis should be performed before initial treatment to exclude malignancy. Fine needle aspiration is suggested rather than core needle biopsy because there is a possibility of hemorrhage due to high vascularity. On the other hand, some prefer core needle biopsy because sufficient tissue can increase diagnostic yield [2].

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As described in this case, large multivaculolated brown fat cells, single central or small eccentric nuclei, large amount of granular cytoplasm, fibrous septae, branching capillaries can be seen in microscopic examination of hibernoma [2].

Furlong et al. suggested four main histological variants of hibernoma: typical, spindle, lipoma-like, and myxoid type. Typical hibernoma and lipoma-like variant are most commonly occurred in thigh, whereas the myxoid and spindle cell type are frequently found in the head and neck region [1]. Typical hibernomas, which is characterized with eosinophilic, pale, and myxoid cells, accounts for about 82% for all cases. The second most common type is the myxoid variant, which has a loose basophilic matrix, accounting for 9% for total cases. Lipoma-like type and spindle cell type are followed, each accounting for 7% and 2% of all cases. Most surgical pathologists do not subclassify aforementioned subtypes because all of them have similar prognosis [2].

Because of its multiple pathological variants and the remaining possibility of misdiagnosis regardless of using clinical, imaging, and pathologic information, some researchers suggested additional chromosomal evaluation to differentiate hibernoma [1].

To date, there is no reported case of malignant transformation or metastasis of hibernoma, so theoretically asymptomatic hibernoma doesn't need treatment [5]. However, due to diagnostic uncertainty of modalities as mentioned above, the treatment of choice is en bloc resection of the mass. En bloc removal is important to prevent recurrence, and also bleeding due to its high vascularity. According to previous articles, almost none of hibernoma patients had recurrence of disease and none died due to the complications of hibernoma with en bloc resection. Some exceptions with recurrences were possibly due to positive resection margins rather than a true recurrence [2].

Since hibernoma is well known for no recurrence after complete removal, the recurrence after the thoracic surgery in this case is also likely to be a result of remnant tumor after the first surgery. Considering the mass extension from thoracic cage into infra-thyroid level at the initial CT, the recurrence might have not been occurred if the primary surgery was planned as co-operation with thoracic surgeon and head and neck surgeon. In addition, even though vocal fold palsy did not occur in this patient, since it was difficult to identify recurrent laryngeal nerve at the second surgery due to the adhesion caused by the first surgery, co-operation should have been preferred as the initial plan for this patient. Even though hibernoma is a benign tumor, this case emphasizes the importance of complete removal of mass to prevent recurrence.

Conclusion

We present a successful removal of recurred hibernoma in neck without any complication. This is a first case report of cervical hibernoma and its management in Korea.

Acknowledgements

None

Funding and Conflicts of Interest

The authors have no other funding, financial relationships, or conflicts of interest to disclose.

Author contributions

Professor Seong Keun Kwon performed surgery, and Young Chul Kim assisted the surgery and took care of the patient during the admission period. Professor Jiwon Koh examined the specimen and pathologically confirmed the diagnosis. Young Chul Kim summarized the case and Jiwon Koh, Seong Keun Kwon read and approved the final manuscript.

Ethical approval

Consent: The study was published with the written consent of the patient's legal guardians.

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Figure Legends

Figure 1: Initial imaging, (A) axial chest CT, (B) coronal chest CT, (C) F-18 FDG PET/CT, (D) ultrasound showing mass from lower neck to posterior upper mediastinum

Figure 2: (A) The lesion was fatty tissue-like yellowsish mass on gross examination. (B) Polygonal cells resembling brown fat with multivacuolated cytoplasm were admixed with mature adipocytes in the background of myxoid stroma. (C) Small, delicate, branching capillaries were noted. (D) No significant nuclear atypism was seen. (E) CD31 immunostaining highlighted hibernoma cells and capillary endothelial cells. (F) The recurred tumor at supraclavicular area had similar histologic features with more pronounced myxoid stroma.

Figure 3: neck CT showing recurred tumor at left supraclavicular area (red arrow), (A) axial view, (B) coronal view

Figure 4: Intraoperational photo of left supraclavicular recurred hibernoma, (A) hibernoma was well circumscribed with capsule, and delicate dissection was performed to save nearby structures, (B) anterior plane of removed hibernoma, (C) posterior plane of removed hibernoma

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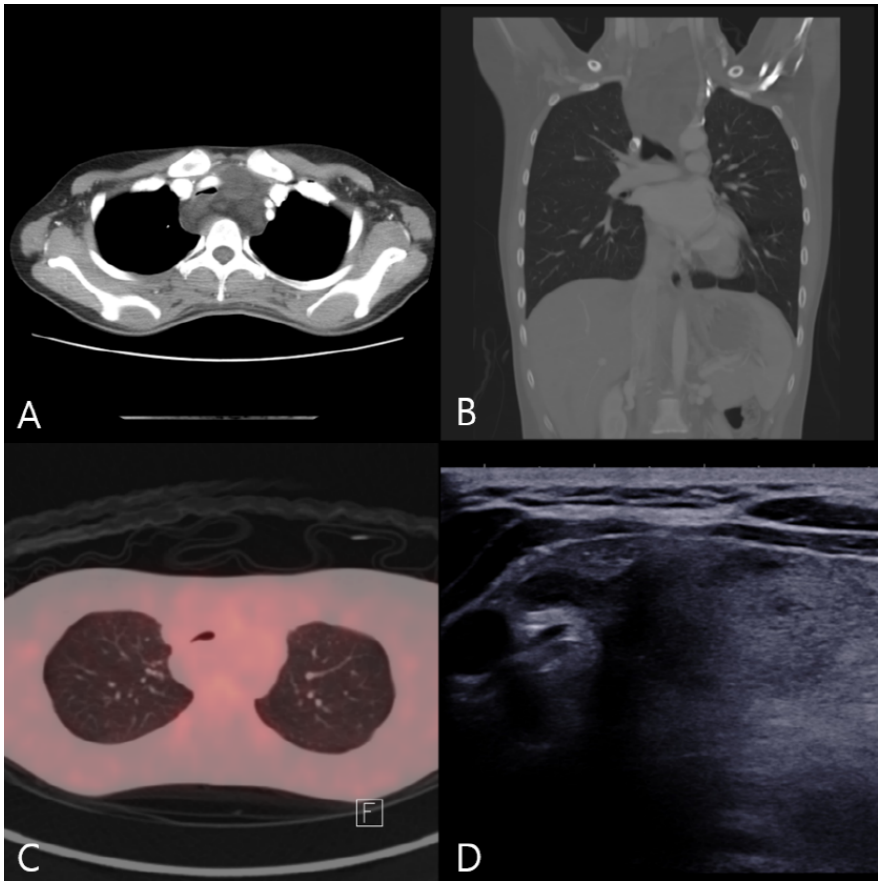

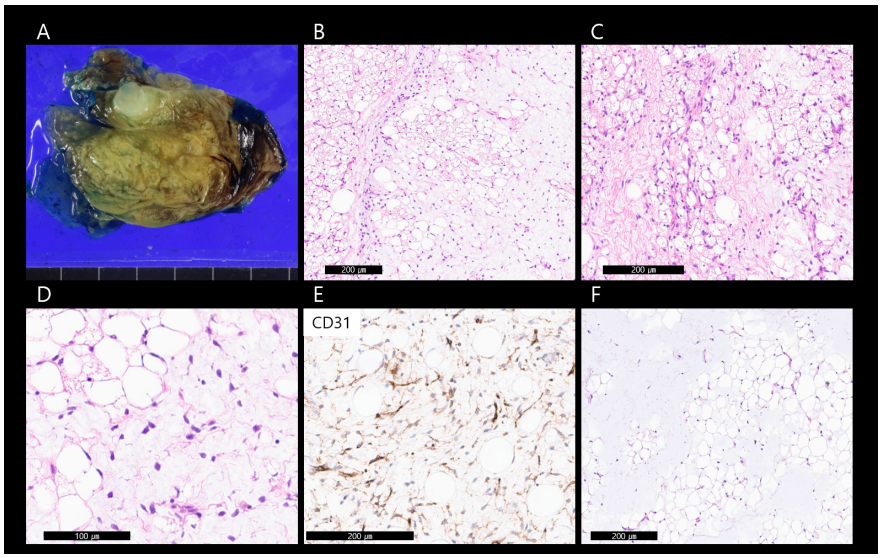

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