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Case Report
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Hibernoma of the Parapharyngeal Space: A Rare Condition and Its Successful Management

*Nalagatla S, Law BZY, Gomati A, Nusky S and Shakeel M**

Department of Otolaryngology-Head & Neck Surgery, Aberdeen Royal Infirmary, Aberdeen, United Kingdom

Abstract

Introduction: Hibernomas are rare, benign soft-tissue tumours composed of cells that are histologically similar to those of brown adipose tissue. In humans, brown fat is typically present during the embryonic period. Its function is to generate heat for thermal regulation (brown fat is also found in hibernating animals, hence the name of the tumour). Hibernoma in the parapharyngeal space is exceedingly rare, clinically present as a bulge in the lateral pharyngeal wall, it is non-painful and usually comes to light incidentally.

Objectives: We aim to present and share our knowledge of dealing with this benign condition focusing on its presentation, investigations and successful resection of parapharyngeal space hibernoma trans-orally.

Methods: A case report and relevant literature review.

Case Report: A 75 year old Caucasian male, otherwise fit and well, presented with a few months' history of a feeling of something stuck in his right throat. He denied any other symptoms. Examination revealed a right parapharyngeal swelling occupying nasopharynx and oropharynx. The computed tomographic scan raised the suspicion of a lipoma. The examination under anaesthesia and incision biopsy carried out through the mouth confirmed the diagnosis of benign hibernoma. On patient's request the lump was completely excised trans-orally. The patient made a smooth recovery. The histological appearances were in keeping with a hibernoma with lipoma-like areas. The patient has remained disease free at 9 years follow-up.

Conclusions: Hibernoma is a slow growing benign tumour of brown fat. It rarely occurs in the head and neck region; presentation as a parapharyngeal mass is extremely rare. However, as demonstrated in our case, it should be included in the differential of a lateral pharyngeal wall tumour. Complete surgical excision is curative. This case also demonstrates the safety, feasibility and efficiency of trans-oral excision of large parapharyngeal space tumours.

Keywords: Hibernoma; Parapharyngeal mass; Trans-oral excision

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Correspondence:
Muhammad Shakeel, Consultant ENT/Thyroid Surgeon, Department of Otolaryngology-Head and Neck Surgery, Aberdeen Royal Infirmary, AB25 2ZN, Aberdeen, United Kingdom.
Tel: *00441224552100*
Fax: *00441224554569*
E-mail: *drshakeel@doctors.org.uk*

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Introduction

Hibernomas are rare, slow-growing benign soft-tissue tumours. They are composed of cells that are histologically similar to those of brown adipose tissue. In humans, brown fat is typically present during the embryonic period of life and has a role in the generation of heat for thermal regulation. The mitochondria in brown fat cells contain a protein, which aids in the uncoupling of electron transport chain regulating heat generation by the cells [1]. Brown adipose tissue is also found in hibernating animals, hence the name of the tumour. Hibernomas arise from areas of brown foetal fat that have remained beyond the embryonic period of life [2]. Hibernomas are more likely to present in those in the fourth or fifth decade of life and are slightly more predominant in the female population [3].

We aim to present and share our knowledge of dealing with this rare benign condition focusing on its presentation, investigations and successful management.

Case Study

A 75-year-old Caucasian male, presented with a few months' history of "something stuck in his right throat". He complained of no other symptoms and was otherwise fit and well. Upon examination a right parapharyngeal swelling occupying nasopharynx and oropharynx with dimensions 8.0x5.5x3.5 cm was seen (Figure 1A and B). A Computed Tomographic (CT) scan was

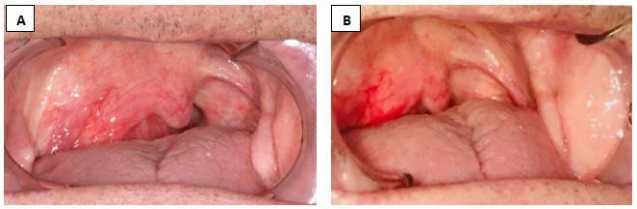

Figure 1: A) Oropharynx showing the right lateral pharyngeal wall swelling pushing the soft palate and uvula to the left and **B)** Oropharynx showing the right lateral pharyngeal wall swelling and a relatively normal looking left lateral pharyngeal wall.

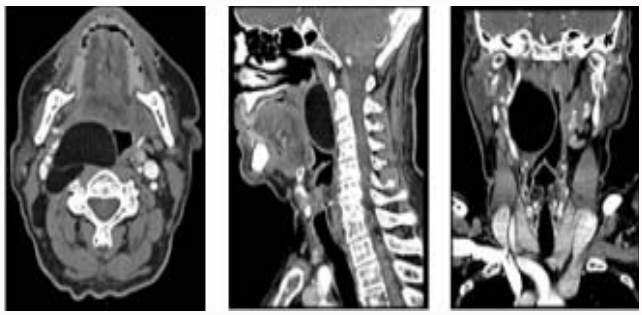

Figure 2: CT scan showing a septum in the right parapharyngeal hibernoma.

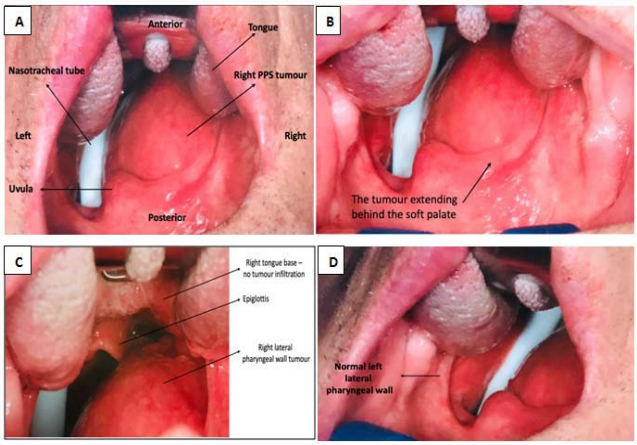

Figure 3: A) The patient is lying supine with Boyle-Davis mouth gag in place. The patient is anaesthetized using a nasal endotracheal tube, **B)** The right lateral pharyngeal wall (right parapharyngeal space - PPS) tumour can be clearly seen extending behind the soft palate into the nasopharynx, **C)** The right PPS tumour is not infiltrating the right tongue base but is touching the epiglottis causing globuspharyngeus and **D)** In comparison the left lateral pharyngeal wall is normal.

performed, which showed a suspected Lipoma occupying the right lateral pharyngeal space (Figure 2).

To confirm the diagnosis the patient underwent examination under anaesthesia (Figure 3A,B,C and D). On palpation the swelling was firm and rubbery with normal overlying mucosa. The swelling was extending from the nasopharynx to oropharynx reaching up to the tongue base without any obvious infiltration. An incisional biopsy was performed through the mouth under general anaesthesia.

The histological appearances were in keeping with a hibernoma with lipoma like areas (Figure 4A and B).

The patient was offered conservative treatment given the benign

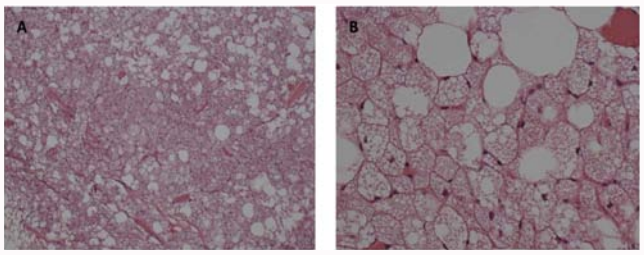

Figure 4: Haematoxylin and eosin stained sections of the resected mass showing typical features of hibernoma. **A)** Original magnification x4- demonstrates an adipose lesion composed of brown fat cells with associated small capillaries and **B)** Original magnification x20-multivacuolated brown fat cells, some with granular cytoplasm; scalloped nuclei with no atypia.

nature of the underlying pathology. However, the swelling was causing significant issues with throat irritation, globus pharyngeus and effortful swallowing. Respecting patient's wishes, the lump was completely excised trans-orally. The technique has been described in an earlier publication [4]. The post-operative recovery was smooth. The patient has remained well and happy 9 years after the operation with no evidence of disease recurrence.

Discussion

Hibernomas are rare benign tumours of brown fat. They can present in the head and neck region as painless lumps. Rarely they do present with other symptoms but cases have reported symptoms of infrequent dysphagia and foreign body sensation in throat [1]. Approximately 200 cases of hibernoma have been reported in the literature to date. The greatest proportion located in the back or thighs and only around 30 cases occurring in the head and neck region [3,5]. Hibernoma in the parapharyngeal space is an exceedingly rare entity. Hibernomas in this location clinically present as a non-painful bulge in the lateral pharyngeal wall, and often come to light incidentally [2]. Successful management of hibernomas include performing appropriate investigations including imaging and carrying out a tissue analysis using needle biopsy or open biopsy. CT imaging, MR imaging and angiography all serve as useful methods in providing information about unknown head and neck masses [2]. However, definitive diagnosis of hibernomas can be achieved by CT-guided biopsy of the mass or by fine needle aspiration cytology [1,6]. Macroscopically hibernomas have a tan to red-brown appearance, which is dependent on the proportion of intracellular lipid present [2]. Histologically, hibernomas differ in the content such as the ratio of multi-vacuolate adipocytes (brown fat) and uni-vacuolate adipocytes (normal fat and lipoma). Furthermore, they vary in the appearance of the polygonal brown fat cells, the associated small capillary proliferation, and the surrounding stromal background. Six histological variants have thus been identified. These include the eosinophilic variant, which is the classical variant containing large numbers of multi-vacuolated adipocytes with abundant, granular cytoplasm and a small, central nucleus. The mixed variant contains a mixture of pale and eosinophilic cells, whilst the pale variant contains only pure pale brown fat cells. These two histological variants are less common. The lipoma-like variant encloses small clusters of brown fat amongst ordinary normal pale fat. The myxoid variant contains only myxoid stroma and the spindle cell variant has thick bundles of collagen fibers, scattered mast cells, and mature adipose tissue [6,7]. Differential diagnoses for hibernomas include lipomas, liposarcomas and rhabdomyomas. One can attempt to differentiate a lipoma from a liposarcomaon the CT scan. Liposarcoma on CT imaging commonly

presents as a non-enhancing low-density mass, with nodules or streaks within the mass [8]. Likewise, liposarcomas and rhabdomyomas have different characteristics on Magnetic Resonance (MR) imaging. MR imaging shows reduced vascularity in liposarcomas and the cytoplasm lacks lipid vacuoles in rhabdomyomas [2]. Curative treatment of a hibernoma involves complete excision of the mass, whilst protecting the important surrounding structures. Due to the high vascular nature of hibernomas, there is a risk of haemorrhage during surgery as well as the risk of postoperative bleeding and haematoma. Care should be taken to avoid these complications [1,6].

Conclusion

A hibernoma in the parapharyngeal space is very rare but an important diagnosis to remember in patients presenting with pharyngeal mass. It is commonly a slow growing tumour which often is asymptomatic but can grow to huge size causing local compressive neck symptoms. Both CT and MRI scans are helpful to investigate this lesion but definite diagnosis is reached by trans-oral or trans-cervical CT scan guided fine needle aspiration cytology. Complete surgical excision is the mainstay of treatment and minimally invasive tans-oral approach is recommended. As of yet, there have been no documented incidents of metastases or malignant transformation associated with hibernomas [2]. Complete trans-oral excision has shown tremendous post-operative prognosis, highlighting this technique as being safe, feasible and effective.

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