CASE REPORTS

UDC: 616-006:617.53
https://doi.org/10.2298/VSP170210077M

Large hibernoma of the neck: A case report

Veliki hibernom vrata

Anton Mikić*†, Miljan Folić*†, Ivan Boričić†‡, Nenad Arsović*†

Clinical Center of Serbia, *Clinic for Otorhinolaryngology and Maxillofacial Surgery, ‡Insitute of Pathology, Belgrade, Serbia; University of Belgrade, †Faculty of Medicine, Belgrade, Serbia

Abstract

Introduction. A hibernoma is a rare benign tumor derived from vestigial remnants of brown adipose tissue. In neonates this tissue makes up about 5% of the body mass and its amount greatly decreases after birth, persisting only in scattered subcutaneous areas. In rare cases, brown fat continues to grow leading to a hibernoma that may be located in the head and neck. We present an illustrative case of a large hibernoma of the neck with infraclavicular extension and discuss about diagnostic and treatment difficulties. **Case report.** A 29-year-old male presented with large, slowly progressive, painless neck mass that was noticed 6 months earlier. Computed tomography (CT) and magnetic resonance (MR) showed a well-vascularized, soft tissue tumor of the lateral region of the neck and supraclavicular fossa with extension below clavicle. Treatment included arterial embolization followed by challenging surgical removal of the tumor. Dissection was performed at III, IV and V levels of the neck, making complete resection possible without the tumor fragmentation or major blood vessels and cranial nerves injuries. The final diagnosis of the hibernoma was made by histopathological analysis. The patient had no signs of recurrence during three-year follow-up. **Conclusion.** Although the CT scan and MR may raise the suspicion, hibernoma is definitely diagnosed by a pathologist. It is very important to exclude the malignant processes, foremost liposarcoma. The tumor fragmentation during surgery should be avoided because the high vascularity of the tumor tissue carries a substantial risk for hemorrhage. Our experience with preoperative embolization and complete tumor resection in this case showed positive impact on the final outcome.

Key words:
diagnosis, differential; embolization, therapeutic; head and neck neoplasms; lipoma; male; otorhinolaryngologic surgical procedures; tomography, x-ray computed.

Apstrakt

Uvod. Hibernom je redak benigni tumor poreklom od zaostataka mrkog masnog tkiva. Kod novorođenčadi ovo tkivo čini oko 5% telesne mase i njegova zastupljenost prilično opada nakon rođenja, zaostajući samo u raštrkanim potkožnim regionima. U retkim slučajevima, mrko masno tkivo nastavlja da raste, formirajući hibernom koji se može naći u regiji glave i vrata. Prikazujemo ilustrativan slučaj velikog hibernoma vrata sa širenjem ispod nivoa klavikule i diskutujemo o dijagnostičkim i terapijskim smernicama. **Prikaz bolesnika.** Na otorinolaringološki pregled se javio 29-godišnji muškarac zbog velikog, spororastućeg, bezbolnog izraštaja na vratu, koji je primetio šest meseci ranije. Kompjuterizovana tomografija (CT) i magnetna rezonanca (MR) pokazali su dobro vaskularizovan, mekotkivni tumor bočne strane vrata i nadključne jame sa širenjem ispod nivoa ključne kosti. Lečenje se sastojalo od arterijske embolizacije tumora, praćene izazovnim hirurškim uklanjanjem tumora. Disekcija je obuhvatila III, IV i V nivo vrata, omogućivši kompletnu resekciju bez fragmentiranja tumora, kao i bez povreda velikih krvnih sudova ili kranijalnih nerava. Konačna dijagnoza hibernoma postavljena je patohistološkom analizom. Tokom trogodišnjeg praćenja bolesnika nije bilo znakova ponovnog nastanka bolesti. **Zaključak.** Iako CT i MR mogu da postave sumnju na hibernom, definitivnu dijagnozu postavlja patolog. Veoma je važno isključiti postojanje maligniteta, najpre liposarkoma. Trebalo bi izbegavati fragmentaciju tumora tokom operacije zato što izrazita vaskularizacija tumora nosi sa sobom rizik od krvarenja. Naše iskustvo sa preoperativnom embolizacijom i kompletnom resekcijom tumora u ovom slučaju je imalo odličan rezultat kao konačni ishod lečenja.

Ključne reči:
dijagnoza, diferencijalna; embolizacija, terapijska; glava i vrat, neoplazme; lipom; muškarci; hirurgija, otorinolaringološka, procedure; tomografija, kompjuterizovana, rendgenska.

Correspondence to: Miljan Folić, Clinical Center of Serbia Clinic for Otorhinolaryngology and Maxillofacial Surgery, Pasterova 2, 11 000 Belgrade, Serbia. E-mail: mfolic@yahoo.com

Introduction

A hibernoma is a rare benign tumor derived from the vestigial remnants of brown adipose tissue. In neonates, this tissue makes up about 5% of the body mass, acting as a kind of a thermal regulator with a high potential to generate heat [1]. Over the first few years, it undergoes rapid degradation, persisting in adults in scattered subcutaneous areas, mostly in the interscapular part of the back [2]. The small depots of brown fat may persist in supraclavicular area of the neck and in rare cases it continues to grow, leading to a benign soft tissue tumor – a hibernoma.

We present an illustrative case of a large hibernoma of the neck with infraclavicular extension and discuss about diagnostic and treatment difficulties.

Case report

A 29-year-old male patient presented with a large, slowly-progressive, painless neck mass that was noticed 6 months earlier. The patient had no neurological deficits and had no problem of swallowing and breathing. A clinical examination showed a soft-tissue tumor, localized in the lower and lateral parts of the neck on the right side, fulfilling the supraclavicular fossa. There was no pathological process found in the pharynx, larynx or nasal cavity. Computed tomography (CT) and magnetic resonance (MR) showed a well-vascularized, soft tissue tumor of the lateral region of the neck and supraclavicular fossa with extension below clavicle that measured 14 × 7 × 4 cm (Figure 1). A few reactive lymph nodes measuring less than 1 cm in the short axis diameter were found close to the internal jugular vein during the ultrasound examination of the neck.

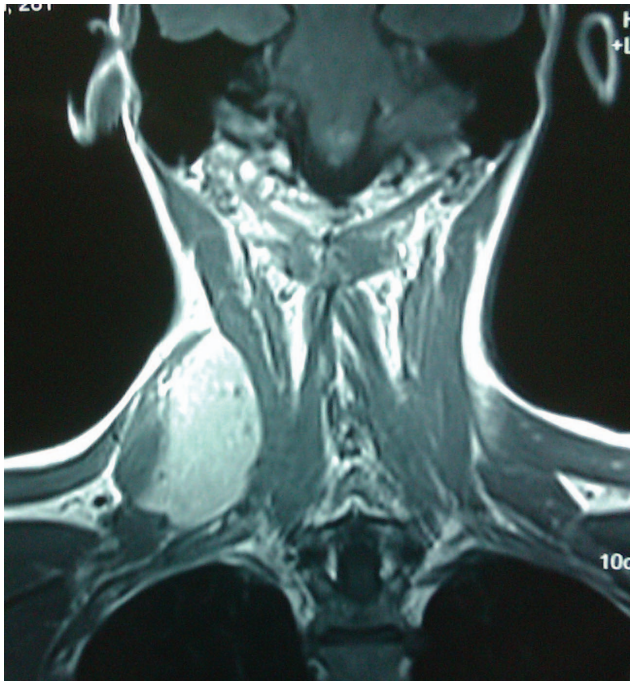

Fig. 1 – Computed tomography (CT) scan of a large neck hibernoma – coronal view.

Pathological vascularization mostly originated from the deep cervical branches of subclavian artery found on the angiographic examination was reduced by selective embolization (Figure 2). In order to make an accurate diagnosis, fine-needle aspiration cytology was performed. Unfortunately, the obtained material was insufficient to make a definite conclusion.

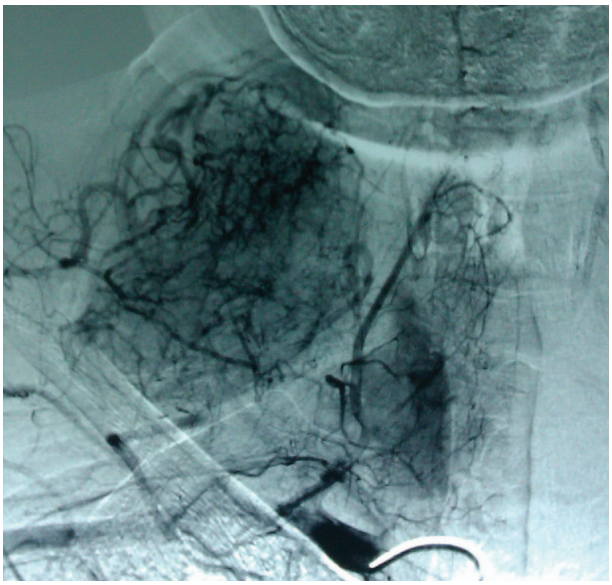

Fig. 2 – Angiographic examination of the right vertebral, subclavian and external carotid arteries.

Embolization was followed by a challenging surgical removal of the tumor. Dissection was performed at III, IV and V levels of the neck, making complete resection possible without the tumor fragmentation, or major blood vessels and cranial nerves injuries (Figure 3).

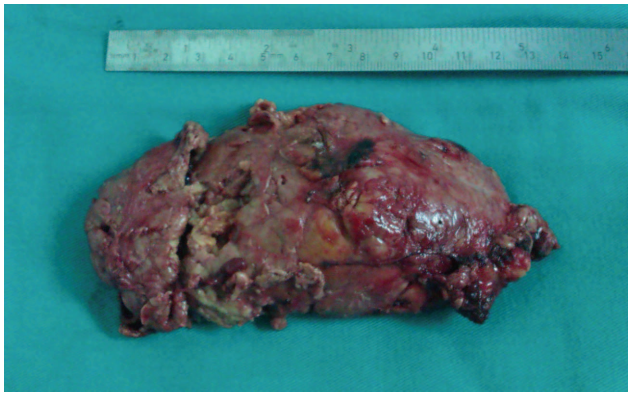

Fig. 3 – Large hibernoma of the neck after complete surgical resection.

A final diagnosis of the hibernoma was made by the histopathological analysis after the surgical resection. The tumor histologically resembles brown fat, consisting of the oval cells with the small centarally placed nuclei without pleomorphism and multivacuolated cytoplasm (Figure 4).

The patient had no signs of recurrence during the three-year follow up.

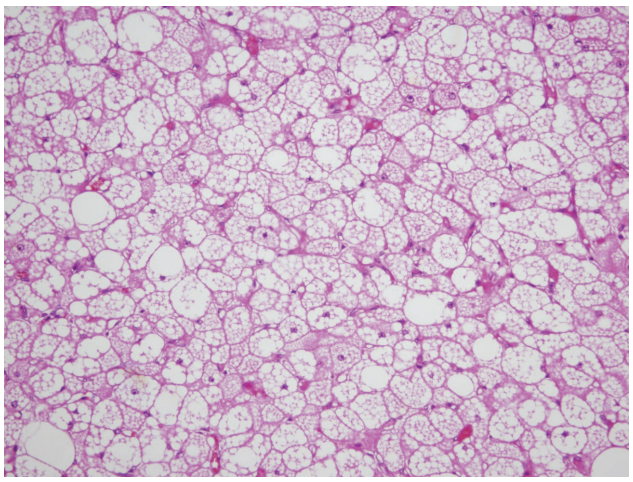

Fig. 4 – Histological appearance of hibernoma [hematoxylin-eosin (HE), ×200].

Discussion

Recent studies using the fluorodeoxyglucose positron emission tomography scanning (PETS) have shown that brown fat remains in very small amounts in adults, which makes a hibernoma, a very rare benign tumor [3] . There are about 15 reports in literature concerning hibernoma of the head and neck which is not as usual area as thigh, shoulder or back. Although it is commonly found in adults, there is a report of the multiple hibernomas in a 1-month-old infant [4]. Data concerning the gender predominance are contradictory, although there is a justified opinion that hibernomas are more frequent in the male population [5].

A hibernoma of the neck usually presents as a slowly-progressive, nontender, mobile mass of a different size, ranging from 5 to 10 cm. Silent growth is the characteristic that makes it diagnosed as a large outgrowth in the soft tissue with no specific symptoms. It was reported that the average period from occurrence to diagnosis of a hibernoma is about 2.5 years [5]. Although some abdominal hibernomas demand an immediate surgical procedure [6], there are no emergency cases of the head and neck hibernoma found in literature.

Considering an important role in nonshivering thermogenesis and high energy demands, the brown fat tissue is made of highly vascularized and mitochondria-rich cells. Therefore, a surgery of the hibernoma is usually associated with intensive intraoperative bleeding. On the other hand, this increased vascularity may help to differentiate hibernomas from other soft-tissue tumors. Some authors recommend arterial embolization to be carried out before the surgery in order to minimize the bleeding that may occur during the intervention [7]. There are no literature evidence that preoperative embolization of a hibernoma affects the surgical outcome, although it may help the surgeon to perform complete resection without the tumor fragmentation [8].

A hibernoma is typically seen on the CT scan as a low-attenuation mass with linear septations. After the contrast administration, enhancement is clearly seen within the septae and more diffusely within the tumor. On a T1- and T2–weighted MR imaging, a hibernoma demonstrates a signal intensity that is increased or approximate to the subcutaneous fat tissue [9]. An increased T1-weighted signal intensity may be seen also in the imaging of other benign on the malignant mesenchymal tumors and specificity is not high enough to make an accurate diagnosis. Diagnostic considerations should include numerous neoplastic lesions, such as lipoma, angiolipoma, hemangioma, hemangiopericytoma, hemangioblastoma, liposarcoma or some types of sarcoma. However, the MR angiography finding of a high T1 signal intensity, combined with a large, curvilinear branching vessels should raise a suspicion of a hibernoma and narrows the differential diagnosis [10].

In suspected cases of a hibernoma with inconclusive radiological findings, a fine-needle aspiration cytology is recommended. A core needle biopsy carries a risk of excessive hemorrhage due to hypervascularity of the tumor and should be avoided, although there are numerous reports considering series of patients with hibernoma that underwent percutaneous biopsy without any complications [11–14].

In cases of the inconclusive cytological findings, the definite diagnosis is made by the postoperative histopathological analysis. There are four histologic types of hibernomas: lobular, myxoid, lipoma-like and spindle cell type of hibernoma. The lobular type is the most common variant of the hibernoma found in 82.4% of the cases [15], including our patient. Oil red O staining is used for the identification of a lipid component and S100 protein positive neoplastic cells are found on immunohistochemistry in about 85% of hibernoma cases [15].

According to their clinical, radiological and pathological characteristics, hibernomas are benign tumors with no potential to cause a fatal outcome. A case report from 1967 by Lowry and Halmos [16] suggested a possible malignant nature of hibernoma due to muscular invasion, although authors had a suspicion about the definite diagnosis and considered a liposarcoma as more accurate pathology finding. In another case of the neck hibernoma, focal infiltration of scalene muscles was not considered as a sign of malignancy, but muscular adherence by the benign tumor? [13].

Furlong et al. [5] reviewed 170 cases of hibernoma and during the follow-up found no metastatic developments or recurrence after complete surgical resection.

Complete surgical resection is considered to be the standard treatment of a hibernoma. Surgeons are recommended to make their best effort to avoid the tumor fragmentation in order to prevent recurrence and minimize the intraoperative hemorrhage. Complete surgical resection provides a good reason to believe that tumor will not reappear, although a long-term follow-up is recommended.

Conclusion

We find this case interesting because it represents an illustrative additional case to a series of rare tumors published so far in literature. The adequate diagnostic and therapeutic approach made a success in treating this massive neck benign tumor. Our experience with arterial embolization followed by complete surgical resection in this case resulted in a complete recovery of the patient.

R E F E R E N C E S

1. *Carter BW, Schucany WG.* Brow adipose tissue in newborn. Proc (BaylUniv Med Cent) 2008;21(3):328-330.
2. *Hui SC, Ko JK, Zhang T, Shi L, Yeung DK, Wang D*, et al. Quantification of brown and white adipose tissue based on Gaussian mixture model using water-fat and T2* MRI in adolescents. J Magn Reson Imaging 2017; 46(3): 758–68.
3. *Pardo JV, Lee JT, Larson RC, Thuras P, Larson AA.* Automated quantification of cold-inducible human brown adipose tissue with FDG PET/CT with application to fibromyalgia. Am J Nucl Med Mol Imaging 2017; 7(1): 24–32.
4. *Baskurt E, Padgett DM, Matsumoto JA.* Multiple hibernomas in a 1-month-old female infant. AJNR Am J Neuroradiol 2004; 25(8): 1443–5.
5. *Furlong MA, Fanburg-Smith JC, Miettinen M.* The morphologic spectrum of hibernoma: A clinocopathologic study of 170 cases. Am J Surg Pathol 2001; 25(6): 809–14.
6. *Sanjuan Rodriguez S, Santamaria Ossorio JI.* Acute abdomen secondary to intra-abdominal hibernoma. Cir Pediatr 2003; 16(3): 152–3. (Spanish)
7. *Kim HS, Lee SG, Son S, Lee K.* Hibernoma in the thoracic back muscle accompanied by neurilemmoma. Korean J Spine 2012; 9(4): 362–4.
8. *Mavrogenis AF, Coll-Mesa L, Drago G, Gambarotti M, Ruggieri P.* Hibernomas: clinicopathological features, diagnosis, and treatment of 17 cases. Orthopedics 2011; 34(11): e755–9.
9. *Tomihama RT, Lindskog DM, Ahrens W, Haims AH.* Hibernoma: a case report demonstrating usefulness of MR angiography in characterizing the tumor. Skeletal Radiol 2007; 36(6): 541–5.
10. *Colville J, Feigin K, Antonescu CR, Panicek DM.* Hibernoma: Report emphasizing large intratumoral vessels and high T1 signal. Skeletal Radiol 2006; 35(7): 547–50.
11. *Lung RJ, Lapidus S, Miller SH, Graham WP.* Hibernoma: report of two cases. J Surg Oncol 1977; 9: 563–6.
12. *Dursun M, Agayev A, Bakir B, Ozger H, Eralp L, Sirvanci M,* et al. CT and MR characteristics of hibernoma: six cases. Clin Imaging 2008; 32(1): 42–7.
13. *Trujillo O, Cui IH, Malone M, Suurna M.* An unusual presentation of a rare benign tumor in the head and neck: A review of hibernomas. Laryngoscope 2015; 125(7): 1656–9.
14. *Guidry CA, McGahren ED, Rodgers BM, Kane BJ.* Pediatric cervicomediastinal hibernoma: a case report. J Pediatr Surg 2013; 48(1): 258–61.
15. *Minni A, Barbaro M, Vitolo D, Filipo R.* Hibernoma of the paraglottic space: an unusual tumour of the larynx. Acta Otorhinolaryngol Ital 2008; 28(3): 141–3.
16. *Lowry WS, Halmos PB.* Malignant tumor of brown fat in patient with Turner's syndrome. Br Med J 1967; 4(5581): 720–1.

Received on February 10, 2017.
Accepted on May 12, 2017.
Online First May, 2017.