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Benign Deep Fibrohistocytoma Of Prevertebral Space: A Rare Occurance

Dr. Nida Baloch, Dr. Sourabh Sharma, Dr Gaurav Bharadwaj,
Dr. Kavita Gupta, Dr. Hani Yousuf

Final Year Pg Resident (Dept. Of Oral And Maxillofacial Surgery), Pacific Dental College And Research Centre, Udaipur
Consultant Oncosurgeon (Dept. Of Surgical Oncology), Pacific Medical College And Hospital, Udaipur
Oral And Maxillofacial Surgeon (Professor And Head) Pacific Dental College And Research Centre, Udaipur
Consultant Pathologist & Professor, Dept. Of Pathology, Pacific Medical College And Hospital
Oral And Maxillofacial Surgeon (Senior Lecturer) At Pacific Dental College And Research Centre, Udaipur

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I. Introduction

The prevertebral space is a potential space between the prevertebral fascia and the underlying vertebral bodies and deep cervical musculature. It extends down the entire length of the vertebral column to the coccyx. However, dense fibrous attachments between the prevertebral fascia and deep cervical muscles tend to contain prevertebral infections and help prevent longitudinal spread.(1) Diseases of these spaces are uncommon but can result in significant morbidity. As these lesions are inaccessible to clinical inspection(2) .Primary lesions include lipoma, liposarcoma, and synovial sarcoma. Direct spread is seen in nasopharyngeal carcinoma, squamous cell carcinoma (supraglottic, oropharyngeal, sinonasal), lymphoma, goiter, chordoma, and primary spinal tumors. Nodal metastasis occurs with squamous cell carcinoma, non-squamous cell carcinoma, lymphoma, papillary thyroid carcinoma, melanoma, and esthesioneuroblastoma. Other conditions include branchial cleft cysts, foregut duplication cysts, ectopic parathyroid adenoma, nerve sheath tumors, vascular and lymphatic malformations, hemangioma, leiomyoma, disk bulge, edema, osteomyelitis, abscess, calcific tendinitis, and tortuous carotid artery. Benign deep fibrohistocytoma (BDFH) is a rare, non-malignant soft tissue tumor composed of fibroblasts and histiocytes. These tumors typically present as slow-growing, painless masses and are predominantly located in deep soft tissues, including the extremities and trunk. Their occurrence in the head and neck region is uncommon. Histologically, BDFH is characterized by a mixture of fibroblasts and histiocytes arranged in a storiform pattern, with limited atypia and mitotic activity. This pattern is similar to that observed in malignant fibrous histiocytoma (MFH), but without the malignant features such as rapid growth, invasion, or metastasis.(3) Patients with BDFH often present with a painless, slowly enlarging mass. While these tumors are benign and rarely metastasize, surgical excision is typically recommended, especially if the tumor causes symptoms or cosmetic concerns.(4) The occurrence of BDFH in the prevertebral space is exceedingly rare, with no documented cases found in current literature. While benign deep fibrohistocytomas are rare, especially in the head and neck region, they should be considered in the differential diagnosis of soft tissue tumors. Their benign nature and characteristic histological features distinguish them from malignant counterparts. Further documentation and study of rare occurrences, such as prevertebral space involvement, are essential to enhance understanding and guide management strategies for these unusual presentations.

II. Clinical Features

Patients with BFH typically present with a slow-growing, painless mass. These tumors are generally well-circumscribed and can vary significantly in size, ranging from 0.5 to 25 centimeters, with a median size of approximately 3 centimeters. While they can occur at any age, the majority of cases are observed in adults, with a median age of 37 years. There is a slight male predominance, with males affected more frequently than females. The most common anatomical locations for BFH are the extremities (58% of cases), followed by the head and neck (22%), trunk (11%), and deep soft tissues such as the retroperitoneum, mediastinum, or pelvis (9%) (5). Histologically, BFH is characterized by bland ovoid to spindle-shaped cells arranged in a storiform pattern, often with admixed lymphocytes. Multinucleate giant cells, osteoclastic giant cells, and foam cells are present in 59% of cases. Other common findings include a hemangiopericytoma-like vascular pattern (42%) and

stromal hyalinization (39%). Treatment typically involves surgical excision, and while local recurrence can occur, metastasis is rare[6].

III. Case Report

A 49 year old female patient reported to our department of oral and maxillofacial surgery PDCRC, Pacific medical university, Udaipur, Rajasthan, India; with complaint of swelling below the mandible and around the neck circumferentially and bilaterally since 2 months with no signs of inflammation. Patient gives history of voice change, dysphagia and dyspnea since the last 2 months. Patient gives no history of prior trauma to head and neck region. Patient was well nourished with sthenic build with normal physical status and or average socio – economic status. The medical , surgical and family histories were unremarkable. On examination the swelling was diffused over the neck bilaterally, ballotable over the lateral sides with no signs of inflammation. The lesion occupied the region from below the chin region anteriorly to clavicular notch and posteriorly from an imaginary line drawn from inferior ear lobe to midclavicular region. Superiorly from inferior border of the mandible to inferiorly upto the clavicle. Further investigation like MRI (head and neck) and FNAC was advised. Patient was admitted to Pacific Medical Hospital, Udaipur for further treatment.

Fig 1. Photograph Showing Marked Swelling Bilaterraly Around The Neck Region

Diffrential Diagnosis

On the basis of clinical and imaging presentation, the following diagnosis were considered- lymphoma, soft tissue sarcoma, nueral/neuroglial tumor, lymphoprolifertaive lesion, dermatofibrosarcoma. Differentiating BFH from other entities is crucial. Malignant fibrous histiocytoma (undifferentiated pleomorphic sarcoma) is a malignant counterpart exhibiting pleomorphism and high mitotic activity. Dermatofibrosarcoma protuberans (DFSP) presents with lower mitotic activity and greater cellularity. Hemangiopericytoma is characterized by a hemangiopericytoma- like vascular pattern. Perineurioma displays thin, elongated spindled cells and is consistently positive for EMA and GLUT1. Plexiform fibrohistiocytic tumor comprises nodules or fascicles arranged in a plexiform pattern. Atypical fibroxanthoma shows severe cellular pleomorphism and atypical mitoses. Dermatofibroma typically presents as a firm, tan-brown nodule with a storiform pattern and factor XIIIa positivity. Angiomatoid fibrous histiocytoma is characterized by cystic blood-filled spaces and histiocyte-like cells[7]. MRI OF NECK (FIG2) reveals well defined lobulated , large T2 STIR hyperintense, heterogenous , solid lesion with its epicenter in prevertebral space. It is etending superiorly in nasopharynx till level of C1 vertebral body. Inferiorly it is extending in the potero- superior mediastinum till D4 vertebral level. The lesion approximately measures 5.1x9.3x17.5cm. The lesion shows marked diffusion restriction with corresponding areas of signal drop on ADC map. The lesion shows predominantly homogenous intense post contrast enchancement. Anteriorly, the lesion is causing mass effect in the form of significant compromise of nasopharyngeal and laryngopharyngeal airway, complete effacement of pyriform fossa. The lesion is diplacing the surrounding vascular structure laterally. The lesion is extending into para pharyngeal space and carotid space on both sides, insinuating in intervening planes. The thyroid gland appears separate from the lesion. The biopsy sections from lesion shows hapzardly arranged cells with elongated to oval bland nuclei. Scatterd lymphocytes seen along with collection of few histocytes. Scattered large cell with abundant eosionophilic cytoplasm and round nuclei seen. Mitosis is rare. Immunohistochemistry study revealed the tumor cell testing positive for CD34 and negative for SS18-SSX, SMA, DESMIN, STAT6, ERG, CD117, DOG1 and MUC4. Ki67 proliferation index was 5%.(FIG 6).

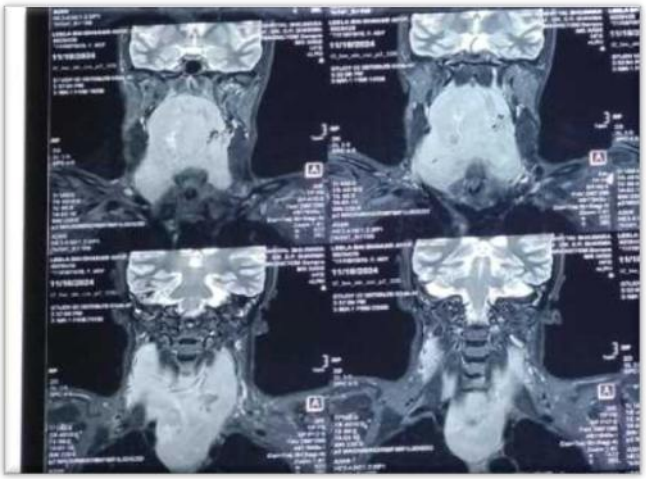

Fig 2. MRI Scan Showing Extension Of Lesion

Treatment

After explaining the diagnosis and surgical procedure to the patient and patient attenders , a written informed consent was obtained from patient. A team of Oncosurgeon and Oral & Maxillofacial surgeon performed the operation under general anesthesia. Under general anesthesia with orotracheal intubation encapsulated excision was performed (FIG3). After adequate haemostasis, lesion was closed in layers. The patient's recovery was uneventful.

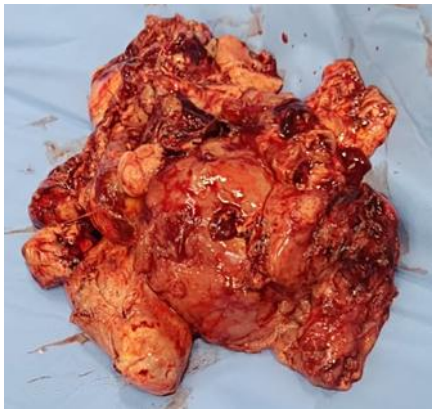

Fig 3. Photograph Showing Excised Mass From Prevertebral Space

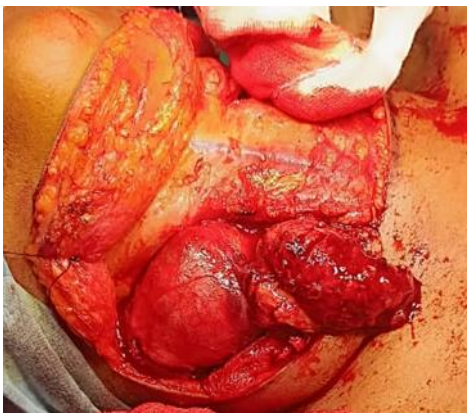

Fig 4. Photograph Showing Mass In The Neck After Exposure

IV. Outcome And Follow Up

On gross examination the excisional tissue consisted of several grey- white to grey brown soft tissue fragments altogether measuring 16x15x6.0cm. Cut surface was grey-white , fleshy along with necrotic and hemorrhagic areas. Histopathological section deomonstrated tumor composed of haphazardly arranged fascicles

of spindle cells with moderate anisoculeosis and variably prominent nucleoli. Additionally, clusters of foamy macrophages and lymphocytes were observed. Areas of coagulative necrosis were seen. Scanty mitotic figures were seen. On the basis of histopathology and ct findings , a final diagnosis of spindle cell neoplasm of fibrohistocytic origin was established. The patient made a full recovery with no neurological deficits.

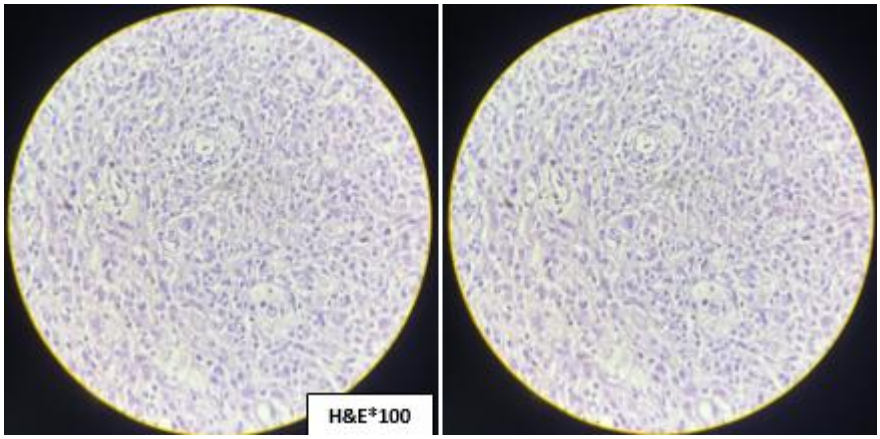

Fig 5. Photomicrograph Showing Histopathological Features

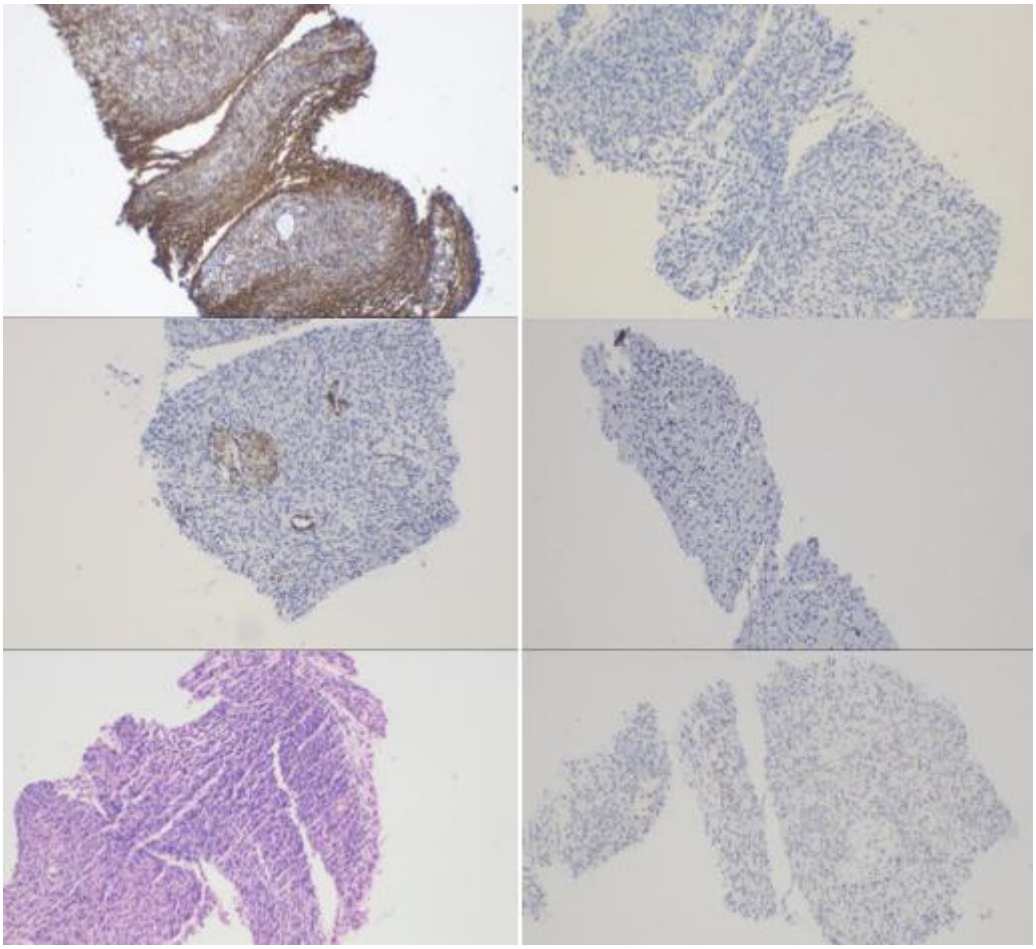

Fig 6. Immunohistochemistry Study Of Sample

V. Discussion

The term "fibrous histiocytoma" was first introduced by American pathologists Shirley L. Kauffman and Arthur Purdy Stout in 1961 to describe a group of mesenchymal tumors exhibiting both fibroblastic and histiocytic differentiation. In 1990, British pathologist Christopher D.M. Fletcher published a series of 21 cases of deep benign fibrous histiocytomas, providing valuable insights into their clinicopathological features.This

early work laid the foundation for understanding the characteristics and behavior of these rare tumors.(8) Typically located in the dermis or subcutaneous tissues, these tumors present as well-circumscribed lesions with a mixed fascicular or storiform growth pattern. The cellular composition predominantly consists of monomorphic spindled or histiocytoid cells, exhibiting limited atypia and mitotic activity(9. The histopathological features of the present case fulfilled the above description. Although the presence of necrosis or its amount hasn't been commented on. In the present case there was a presence of a huge amount of necrosis present which is not very common in these type of lesions. In the present case the presence of Ischemic type of necrosis was ascribed to the fact that the lesion had grown remarkably in size which resulted in compromise of vascularity. Thus, promoting Ischemic necrosis. Studies have shown that none of the benign fibrous histiocytomas expressed CD34. However, in deep fibrous histiocytomas, CD34 positivity is more prevalent, with about 40% of cases showing peripheral staining, typically weak and patchy. This pattern contrasts with dermatofibrosarcoma protuberans, a malignant counterpart, which exhibits strong and diffuse CD34 positivity throughout the lesion. Therefore, while CD34 is generally negative in superficial BFH, its expression in deeper variants can aid in distinguishing them from malignant tumors(10). Our patient in this case presented CD34 test positive. Deep benign fibrous histiocytomas (BFHs) of the head and neck are rare tumors that can be challenging to differentiate from other soft tissue lesions due to overlapping clinical and histopathological features. A comprehensive differential diagnosis includes malignant fibrous histiocytoma (undifferentiated pleomorphic sarcoma), dermatofibrosarcoma protuberans, angiomatoid fibrous histiocytoma, atypical fibroxanthoma, solitary fibrous tumor, and collagenous fibroma (desmoplastic fibroblastoma). A thorough histopathological examination, supported by immunohistochemical staining and clinical correlation, is essential for accurate differentiation, ensuring appropriate management and prognosis.(11)

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