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CASE REPORT

Tumefactive fibroinflammatory lesion: A rare etiology for a neck mass in a child

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Summary Tumefactive fibroinflammatory lesions are rare fibrosclerosing disorders that can present in the head and neck. Clinically, their infiltrative and destructive nature would suggest an aggressive malignancy. However, histologically, they are benign and do not metastasize. We present a case of a tumefactive fibroinflammatory lesion in a 4-year old, the youngest patient reported, with a review of this disease entity and discuss its treatment options.

1. Introduction

First described by Rice et al. [1] and originally called ''sclerosing cervicitis'', tumefactive fibroinflammatory lesions (TFLs) of the head and neck were later termed as such by Wold and Weiland [2]. These are infiltrative fibrosclerosing processes with a locally destructive nature that resembles a sarcoma. However, they are histologically benign and do not metastasize. The etiology of these lesions has not been identified. TFLs appear similar histopathologically to other idiopathic fibrosclerosing lesions including Riedel's thyroiditis, orbital inflammatory pseudotumor, sclerosing mediastinitis, sclerosing retroperitonitis and sclerosing cholangitis. TFLs are rare entities with only 26 cases reported in the literature with a vast majority of cases occurring in the adult population. We present a case of a TFL occurring in a 4-year old, discuss the clinical, radiologic, and pathologic characteristics of the lesion, and review the management strategy.

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2. Case report

A 4-year-old girl presented with an enlarging mass in the left anterior neck. One-day prior, the mass was noted by the patient's mother and reported to have grown in size "overnight". The patient was otherwise healthy with no other pertinent past medical history. Physical examination revealed an afebrile, well-appearing young girl with a 5 cm × 3 cm × 2 cm mass left of midline in the anterior neck, with mild overlying erythema. The mass was firm, mobile and non-tender. Adjacent lymph nodes were small and cranial nerve functions were all grossly intact.

A neck ultrasound revealed a non-cystic, ill-defined soft tissue mass in the left neck, with increased flow on Doppler. Computed tomography (CT) of the neck demonstrated a 3 cm × 3 cm heterogeneous, contrast-enhancing, soft tissue mass involving the entire left thyroid lobe and extending superiorly to the level of the hyoid with anterior displacement of the sternocleidomastoid muscle and deviation of the trachea to the right (Fig. 1a and b). The CT also showed a few mildly enlarged lymph nodes and no fluid collections; bone windows showed no lytic or blastic osseous lesions. Bloodwork showed an initial leukocyte count of 11.4×10^3/mcl, normal electrolyte panel, normal thyroid function, normal calcitonin and PTH levels. Further testing was negative for EBV, Histoplasma, Blastomycosis, Francisella, Bartonella, and CMV. Tuberculin skin testing and blood cultures were negative.

The day after presentation, the patient underwent direct laryngoscopy and incisional biopsy of the mass. Intraoperatively, no opening was identified in the left piriform sinus to suggest a branchial cleft anomaly. Upon incision, the mass was noted to be firm and caramel-colored. The initial pathology report was non-diagnostic, and was read as a "reactive fibroinflammatory process". A repeat incisional biopsy was performed 2 weeks later, at which time the patient demonstrated a persistent mass and progressive erythema of the overlying neck skin. Most of this mass and the involved skin were excised. Histopathology of this second biopsy was consistent with a tumefactive fibroinflammatory lesion. The lesion was characterized by a paucicellular proliferation of bland fibroblasts with dense collagenization, accompanied by a patchy inflammatory infiltrate composed of lymphocytes, plasma cells and eosinophils (Fig. 2a). The fibroinflammatory process extended into skeletal muscle (Fig. 2b).

Subsequently, the patient underwent evaluation for other forms of fibrosclerosing lesions with CT scans of the chest, abdomen and pelvis. These revealed no additional lesions. Her liver function tests did not indicate sclerosing cholangitis, and ophthalmological evaluation showed no evidence of orbital pseudotumor. Treatment with steroids was planned, but the lesion slowly decreased in size spontaneously until it was no longer clinically evident. A repeat neck CT 4 months after initial presentation showed no evidence of residual disease.

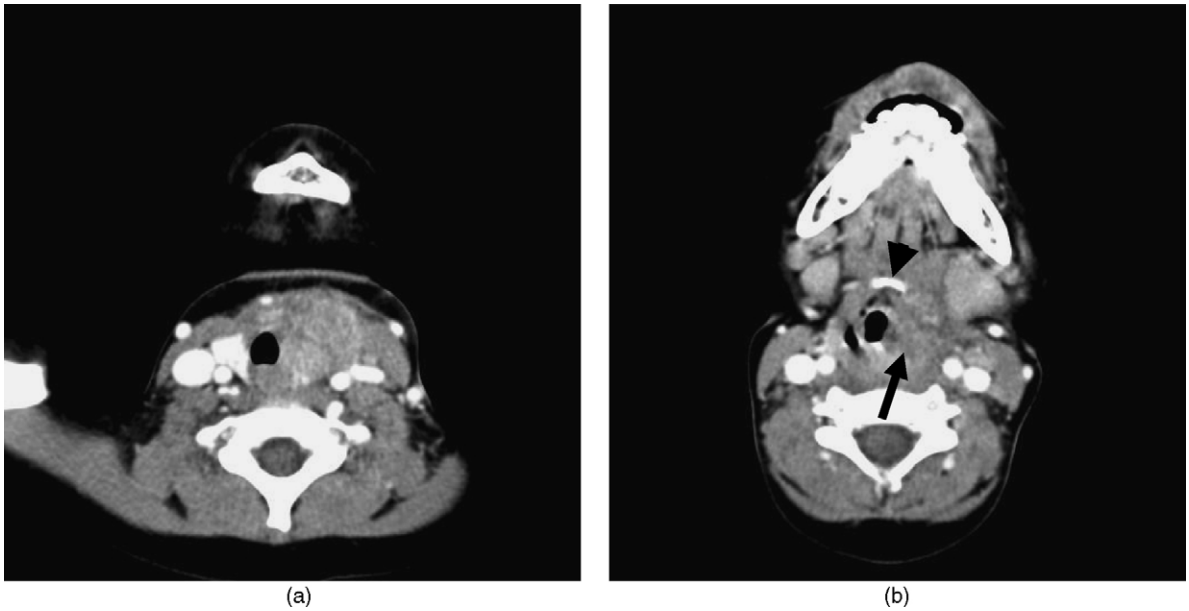

Fig. 1 (a) Axial neck CT scan with contrast showing 3 cm × 3 cm enhancing soft tissue mass in anterior left neck involving left thyroid lobe with mass effect displacement of left sternocleidomastoid muscle and trachea, and with infiltration of adjacent subcutaneous fat. (b) Axial neck CT scan with contrast showing superior extent of mass (arrow) at the level of the hyoid bone (arrow head).