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Case Report

Primary cervical leiomyoma: A rare cause of a posterior neck mass in a pediatric patient

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ABSTRACT

A 13-year-old male presents for evaluation of a right-sided posterolateral neck mass, first noted four years prior to presentation; incisional biopsy two years ago suggested a benign lymph node. Recent growth and increased pain prompted referral to our tertiary care center. MR imaging revealed a densely calcified mass in the right posterior paraspinous muscles with intense enhancement with gadolinium contrast, approximately 5 cm × 2.8 cm x 4.6 cm. Incisional biopsy showed leiomyoma with extensive dystrophic calcifications. This case describes a rare finding of extraesophageal leiomyoma of the neck; this is only the second such case reported in a pediatric patient.

1. Introduction

Leiomyomas are benign neoplasms of smooth muscle origin, most commonly associated with uterine myometrium, with much lower incidence in the gastrointestinal tract, skin and lower extremities. Some of the proposed mechanisms for the development of leiomyoma include a congenital origin, blood flow disturbance, infection, and the involvement of estrogen [1]. These tumors occur most often in middle-aged women, possibly due to progesterone receptors on the benign mass [2].

Less than 1% of all leiomyomas occur in the head and neck, as this region contains relatively little smooth muscle [3]. In these rare cases, tumor origin most often appears to be esophageal [4]. Extra-esophageal head and neck leiomyomas are exceedingly rare, particularly in the pediatric population [4]. The histogenesis of leiomyomas remains a point of debate; however the most prominent theories for origin of development remain vascular smooth muscle, the main component of the wall of small blood vessels, and multipotent mesenchymal cells [5,6].

2. Case report

A 13-year old male presented to our institution with a right posterolateral neck mass, which he reported had been present for several years. He denied odynophagia or dysphagia. He also did not complain of any subjective facial weakness, vertiginous symptoms, or motor coordination disturbances. Incisional biopsy two years earlier had suggested a reactive lymph node. However, the mass had progressively enlarged and become more painful. Physical examination demonstrated a very firm mass deep to the posterior neck musculature and fixed to the surrounding soft tissues. Initial radiological impression indicated a large heterogeneous and highly calcified mass either compressing or slightly invading the right vertebral artery, with potential erosion of the adjacent calvarium (see Fig. 1). Incisional biopsy was performed. A large, 5-cm, well-circumscribed, pale yellow mass with large, granular intra-lesional calcifications was found in the right posterior neck deep to the trapezius muscle. Internally, the mass had a crumbly appearance and crunchy texture due to the calcifications, which made incision challenging. The biopsy demonstrated a leiomyoma (see Fig. 2). Complete resection of the mass took place six weeks later (see Fig. 3). The superior aspect of the mass was adjacent to the skull base. Despite the imaging appearance, no major vasculature or cranial nerves were directly encountered, though the vertebral artery was clearly palpable and separated from the mass by a thin layer of tissue. The postoperative course has been unremarkable. The patient was last seen one month postoperatively and was doing well. Further follow-up was planned on an as-needed basis, and he has thus far not re-presented to our clinic.

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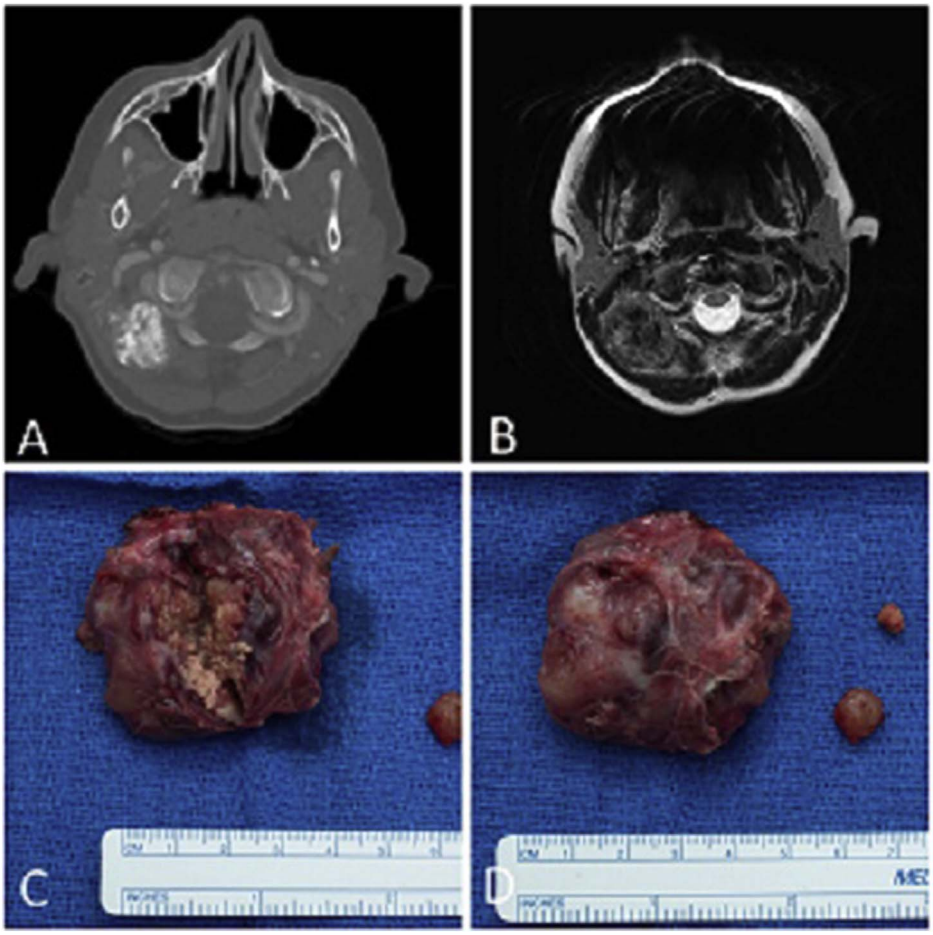

Fig. 1. Axial imaging demonstrating calcified mass abutting the left vertebral artery with (A) CT and (B) T2 MR; appearance of the mass after excision showing (C) obvious granular calcifications and (D) external lobulation.

3. Discussion

The three main types of leiomyomas typically found in the head and neck include: vascular (also known as angioleiomyoma or angiomyoma), primary (non-metastatic), and intranodal (spindle cell lesion of lymph node) [3]. The most common sites of leiomyoma in the head and neck region include the lips (27.46%), tongue (18.30%), cheeks and palate (15.49%), gingiva (8.45%) and mandible (5.63%), and the remainder of these (24.67%) appear to arise from the esophagus or pharynx [3,4]. The differential diagnosis for a fibroid mass of smooth muscle origin with intra-lesional calcification in the pediatric population is quite broad (see Table 1). Our patient had no history of animal exposures, mycobacterial exposures, or other symptoms to suggest leukemia or lymphoma and no physical exam findings to suggest lymphoma.

While leiomyomas often contain calcifications, primary, non-metastatic neck lesions with calcification and ossification are extremely rare, and cannot be presumed to be benign [1]. Clinical manifestation of leiomyoma tends to be an otherwise asymptomatic, slow-growing mass [1]. A characteristic histopathological finding of leiomyoma is proliferating spindle cells in a tangled arrangement [1]. Tumors of smooth muscle origin, including leiomyosarcoma, have been characterized through the use of immunohistochemistry by the presence of α-SMA and h-caldesmon, very highly specific myogenic markers for smooth muscle involvement [7,8].

Histological features of the mass can be distinguishing to the point of accurate diagnosis. The lack of amianthoid fibers, interstitial hemorrhage, intracytoplasmic actin-positive globular inclusions, and prominent nuclear palisading would rule out IPM from the differential [9]. The degree of angiogenesis in the smooth muscle neoplasm can often distinguish leiomyoma from angiomyoma (vascular leiomyoma), and epithelioid leiomyoma (leiomyoblastoma), which tend to be more vascular and less common than the solid form of leiomyoma [1]. Determining leiomyoma from leiomyosarcoma preoperatively can be challenging, and indeed our case had significant concerns about vertebral artery and calvarial invasion on preoperative imaging. We recommend incisional biopsy of the lesion to obtain a large tissue sample, and to definitively identify relevant histopathological features. Needle biopsy may also be an option, though the extremely hard, granular contents of the tumor might make this approach challenging.

The mechanism underlying intra-lesional calcification with leiomyoma is unknown. Some studies have demonstrated increased intracellular calcification through circulatory disturbances which arise in tumor tissue, as well as hyaline degeneration in tumor tissue and subsequent calcification due to the abnormally high percentage of collagen fibers present [10].

4. Conclusion

Primary cervical (extraesophageal) leiomyoma is extremely rare, and has been reported only once before in a pediatric patient. Differentiation of this lesion from other neck masses is difficult prior to resection, as demonstrated by the broad differential diagnosis for a fibroid mass of smooth muscle origin with intra-lesional calcification. Surgical resection of leiomyomas is the currently accepted course of treatment based on the limited literature available; the prognosis is excellent, with recurrences being rare.