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A Child with Internal Jugular Vein Phlebectasia Masquerade as Cervical Lymphadenopathy

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Abstract

Pediatric neck swelling is common; the most common pathology encountered in primary care is benign reactive cervical lymphadenopathy. Internal jugular phlebectasia is rare and may be missed on initial presentation. The Valsalva manoeuvre is simple yet clinically useful, narrowing the differential diagnosis. Imaging studies, including Doppler ultrasound and contrast-enhanced computed tomography, are the first-line modalities to confirm the diagnosis and rule out other diagnoses. Management is primarily conservative, with surveillance follow-up, while surgery is reserved for symptomatic cases or for cases with complications.

Keywords: Internal jugular vein phlebectasia, cervical lymphadenopathy, Valsalva manoeuvre

Introduction

Neck swelling is common in the pediatric population and can be classified as congenital, inflammatory, and neoplastic pathologies. Benign reactive cervical lymphadenopathy is the most common lesion seen in primary care, while other unusual pathologies like congenital anomalies, neoplasm and vascular malformations are more commonly seen at the otolaryngology clinic[1]. Some of the lesions could be discovered as incidental findings during head and neck examinations and may require additional manoeuvres to elicit the physical signs like Valsalva manoeuvre.

Neck swelling aggravated by the Valsalva manoeuvre in a child is relatively rare, and the differential diagnoses could be internal jugular vein (IJV) phlebectasia, laryngocele, or herniation of a superior mediastinal mass[2]. IJV phlebectasia is a benign anatomical variant of IJV of unknown aetiology, characterised by non-tortuous dilatation of IJV[3]. Although rare, differentiating this from other, more common pathologies such as cervical lymphadenopathy is paramount because the management is markedly different.

Case Report

Informed consent was obtained from both parents for case write up including permission for publication of all photographs and image. A 7-year-old boy with no known medical illnesses presented with a painless swelling of the right anterior neck for three months. He initially presented to primary care with an upper respiratory tract infection and

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acute tonsillitis, and the neck swelling was discovered during the neck examination. A diagnosis of acute tonsillitis with cervical lymphadenitis was made, and he was prescribed a course of oral antibiotics for one week. Although the other symptoms were completely resolved after two weeks, the neck swelling persisted. The tuberculosis workup and other blood investigations were unremarkable, and he was referred to our tertiary centre for further management.

On further questioning, he denied a history of trauma to the neck, prolonged cough, fever, dysphagia, odynophagia, or changes in voice. There were no constitutional symptoms, no hyperthyroid or hypothyroid symptoms, and no family history of malignancy. The patient and his parents did not notice any aggravating or relieving factors.

On examination, there was a diffuse swelling of the right anterior neck, extending from the angle of the mandible to the level of the cricoid cartilage (**Figure 1A**). The overlying skin was normal, non-pulsatile, firm in consistency, non-tender, immobile and did not fix to the overlying skin. The swelling did not move with deglutition but increased in size during the Valsalva manoeuvre (**Figure 1B**). Other head and neck examinations, including flexible nasopharyngolaryngoscopy and cranial nerve examinations, were unremarkable.

Computed tomography scan of the neck revealed right IJV was relatively prominent compared to the left side, with a maximal diameter of 2.4 cm (**Figure 2**). The diameter of the left IJV was 0.8 cm. No filling defects were seen bilaterally. The other structures of the neck, particularly laryngeal subsites, were unremarkable.

A diagnosis of IJV phlebectasia was made; since the patient was asymptomatic and had no clinical or radiological evidence of thrombosis, he was managed conservatively with regular follow-up.

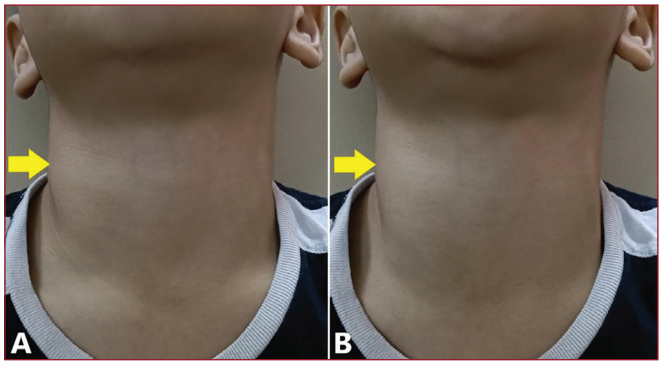

Figure 1. A diffuse swelling at the right anterior neck, extending from the angle of the mandible to the cricoid cartilage level (A), increasing in size on the Valsalva manoeuvre (B)

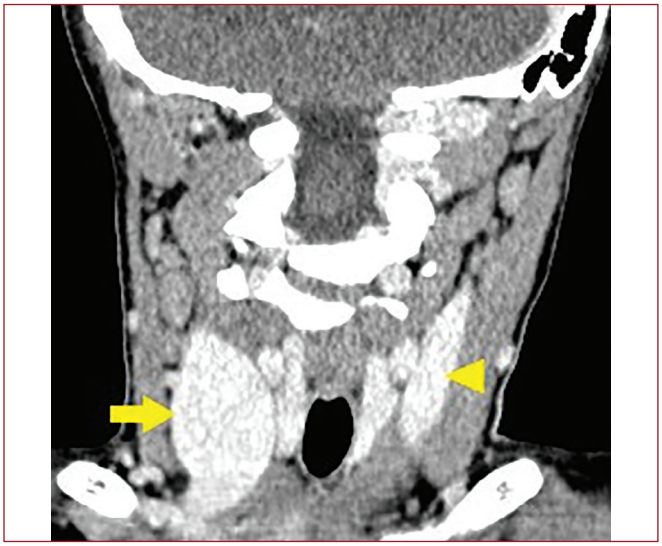

Figure 2. Contrast-enhanced computed tomography shows right IJV (arrow) is relatively prominent compared to the left side (arrowhead), with a maximal diameter of 2.4 cm

IJV: Internal jugular vein

Discussion

IJV phlebectasia was first described by Harris as a blood-filled cyst in 1928[4], however the first definitive case report was published in 1929 by Zukschwerdt[5], and thereafter further characterised by Gerwig in 1952[6]. It is a rare type of vascular anomaly and is characterised by non-tortuous dilatation of the vein, which is different from the term varix, which refers to tortuous dilatation[7]. A systematic review by Figueroa-Sanchez et al.[3], including both pediatric and adult, found 97 original articles, with a total of 247 cases; 206 cases were younger than 18 years. Another, more recent, systematic review by Alkusayer et al.[8], which involved only pediatric cases, found 51 articles with a total of 169 cases.

The predominant symptom is painless neck swelling, while other symptoms are relatively rare, such as neck pain, voice changes, shortness of breath, dysphagia, and Horner's syndrome[3,8]. Incidental finding during neck examination is reported in 22% of cases[3]. This could be due to the painless small lesion, or the patient has a relatively thick neck, resulting in less obvious swelling, which is not of concern to the patient. In addition, the swelling could be intermittent and visualised only during straining or while performing the Valsalva manoeuvre. The classical sign is soft swelling on palpation, which, in more than 90% of cases, increases in size with the Valsalva manoeuvre[3,8].

In our case, neck swelling was found incidentally on examination when the patient presented with other complaints. It was initially diagnosed with cervical lymphadenopathy because the patient had a concomitant upper respiratory tract infection and acute tonsillitis. Without performing the Valsalva manoeuvre to elicit the sign or obtaining a detailed history of straining that aggravates the neck swelling, the diagnosis of IJV phlebectasia could be missed. Furthermore, the neck swelling was firm rather than soft, as typically described in the literature. This clinical finding can be explained by the position of the IJV, which lies medial and posterior to the sternocleidomastoid muscle.

The differential diagnosis in pediatrics differs from that in adults. Laryngocele and superior mediastinal tumour/cysts are the main differential diagnosis in pediatric patients, while Zenker's diverticulum and pharyngocoeles could be added as another differential diagnosis in adult[2,8,9]. Apart from the IJV, other less common neck veins involved are the external and anterior jugular veins[7]. All these lesions become prominent with the Valsalva manoeuvre. Branchial cyst and cystic hygroma are other differential diagnoses, especially in

cases of neck swelling that does not change in size with increased intrathoracic pressure.

The aetiology remains incompletely understood owing to the rarity of the disease. Some authors proposed trauma as the leading cause, such as direct injuries to the neck, central venous catheterisation, positive-pressure ventilation and neck surgeries[3]. Other proposed theories are congenital origin, abnormal IJV reduplication, increased scalenus anticus muscle tone, and compression of the vein between the head of the clavicle and the cupula of the right lung[8]. Another interesting theory proposed by LaMonte to explain the right-sided predominance of the lesion is the proximity of the right innominate vein to the right apical pleura, which transmits intrathoracic pressure to the right IJV[3].

Ultrasound with Doppler while the patient is performing the Valsalva manoeuvre is the preferred imaging study[10]. It is radiation-free and readily available with a short waiting list, but is highly operator-dependent and less effective at delineating deeper structures. Eksioglu et al.[11] suggest an anteroposterior diameter of more than 15mm as the cut-off point to diagnose IJV phlebectasia based on ultrasound measurement. Another commonly employed imaging study is contrast-enhanced computed tomography (CECT), as it provides better definition of the lesion and its surrounding structures. In the present case, we chose CECT as the imaging modality because we considered it provided better information about the lesion and helped rule out other differential diagnoses.

The complications such as thrombosis and Horner's syndrome are rare, while spontaneous rupture is never reported[7]. Conservative management is preferred and safe, whereas surgery should be reserved for the symptomatic cases or in the presence of complications[3,7,10]. Although it rarely causes significant problems and may regress on its own, surveillance follow-up is recommended to observe for any complication[7].

Conclusion

IJV phlebectasia is much less common than cervical lymphadenopathy. Medical practitioners, especially in primary care, should be aware of this rare disease entity. Detailed history and examinations, including the Valsalva manoeuvre, are paramount. An appropriate imaging study should be employed as the first-line modality to confirm the diagnosis and rule out other diagnoses in the differential.

Ethics

Informed Consent: Informed consent was obtained from both parents for case write up including permission for publication of all photographs and image.

Footnotes

Author Contributions: V Sha Kri ED: Surgical and Medical Practices, Concept, Design, Data Collection or Processing, Analysis or Interpretation, Literature Search, Writing.

Conflict of Interest: The author has no conflicts of interest to declare.

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