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Dysphagia because of unilateral internal jugular vein phlebectasia in an infant



Jegadeesh Sundaram ^a, Prema Menon ^{a,*}, Shyam K.S. Thingnum ^b, Katragadda Lakshmi Narasimha Rao ^a

- ^a Department of Pediatric Surgery, Postgraduate Institute of Medical Education and Research, Chandigarh, India
- ^b Department of Cardiothoracic Surgery, Postgraduate Institute of Medical Education and Research, Chandigarh, India

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ABSTRACT

Phlebectasia affecting the internal jugular vein is a rare cause of a benign neck swelling in children. They are mostly asymptomatic and therefore managed conservatively. Ligation of the vein and excision is usually avoided owing to the worry of raised intracranial pressure. We report a case of a large right internal jugular vein phlebectasia, causing dysphagia in a 7 month old male child. Contrast enhanced computed tomography with 3-D reconstruction helped in pre-operative anatomical delineation, especially of the lower extent. It was excised through a cervico-thoracic approach with postoperative amelioration of symptoms. Contrary to expectation, the ectasia was not friable and was covered with a pseudocapsule of fibrofatty tissue, making excision easy.

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Cervical phlebectasias are rare differential diagnosis of neck swellings in children and can affect the internal, external or anterior jugular vein [1]. They are usually asymptomatic and tend to be managed conservatively. Surgery may also be avoided owing to worries of intracerebral edema on ligating the internal jugular vein (IJV) [2]. Cosmetic and psychological effect on the family should also be considered during decision making because of the location. We report a case of an isolated right internal jugular vein phlebectasia (IJVP), presenting with dysphagia, with amelioration of symptoms after surgical excision.

1. Case report

A 7 month old boy, weighing 6.1 kg, was referred with a 3 months history of dysphagia, choking while taking feeds, followed by irritable cry and a swelling appearing on the right side of the neck. A nontender, 4×3.5 cm, soft, cystic swelling posterior to the lower one third of the right sternocleidomastoid muscle, extending to the anterior and posterior triangle of the neck appeared only when the child cried. On auscultation, there was no bruit or venous hum over the swelling. There were no clinical features suggestive of connective tissue disorder or other vascular anomalies. Apart from a Doppler ultrasound (Fig. 1 A), contrast enhanced computed tomography (CECT) of the neck and chest (Fig. 1 B) with 3-dimensional (3D) (Fig. 2) reconstruction was

E-mail address: menonprema@hotmail.com (P. Menon).

performed. The 2-D echocardiogram was normal. The phlebectasia was expected to be friable and in view of its extension below the neck, an upper median sternotomy was initially performed for control of the distal IJV above its junction with the right subclavian vein. It was then extended into the neck through a cervical skin crease incision. The ectatic segment was found to be completely surrounded by a pseudocapsule of fibro-fatty tissue and was not friable. (Fig. 3) It was excised in toto after ligation of both ends. (Fig. 4 A & B). Postoperatively, the child had transient increase in blood pressure which resolved spontaneously within 2 days. Histopathological examination revealed the muscle layer to be replaced by fibrosis and myxoid degeneration, congested capillaries, undulated internal elastic lamina and flattened endothelial lining. At a follow up of 9 months, he remains asymptomatic with parental satisfaction and weight gain.

2. Discussion

Phlebectasia, also known as venous congenital cyst, venous aneurysm, venous ectasia and essential venous dilatation depicts an abnormal dilatation of a vein without tortuosity [3]. A varix, on the other hand, implies both dilatation and tortuosity. An IJVP is a rare cause of a non-tender, compressible swelling in the neck, characteristically becoming more prominent on Valsalva maneuver [4]. They are usually asymptomatic although an uncomfortable feeling while swallowing, giddiness, change in voice etc. have been noted [1–5]. These could be attributed to the pressure effect on neighboring structures such as the esophagus, the cervical sympathetic nervous system as well as the lower cranial nerves within the carotid sheath, especially

 $^{^{*}}$ Corresponding author at: Room No. 3103, Level 3-A, Department of Pediatric Surgery, Advanced Pediatric Centre, P.G.I.M.E.R., Chandigarh 160012, India. Tel.: $+91\,172\,2747585x5320$; fax: $+91\,172\,2744401$, +912745078.

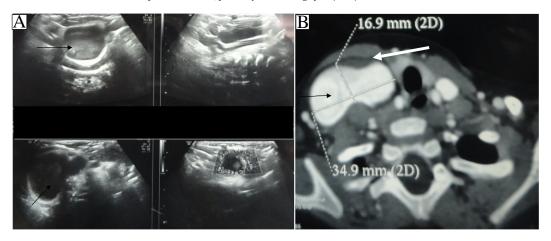


Fig. 1. (A) Doppler ultrasound and (B) contrast enhanced computed tomography (CECT) of the neck showing right internal jugular vein phlebectasia (black arrow) with pseudocapsule (white arrow).

the vagus nerve including its recurrent laryngeal nerve branch. The index case presented with dysphagia and choking which resolved after excision.

The first report of IJVP is attributed to Gruber in 1875 and later on, in the English language literature to Harris [1,6,7]. Forty-five more cases were reported by 1997 [8]. We came across 103 more cases, making it

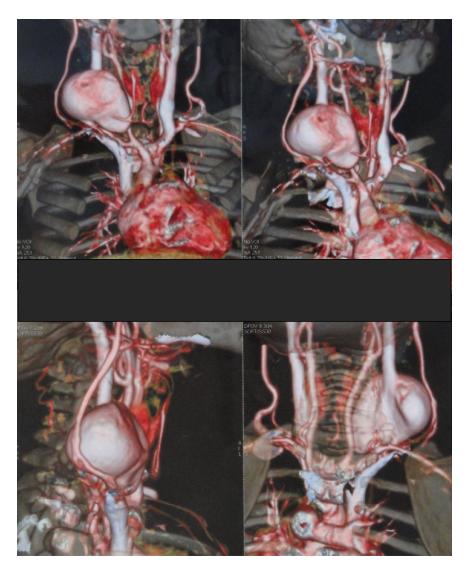


Fig. 2. Contrast enhanced computed tomography (CECT) of the neck and the chest with 3-dimensional reconstruction, shows a $25.5 \times 34.3 \times 26.6$ mm (anteroposterior \times transverse \times craniocaudal) fusiform dilatation of the right internal jugular vein ending approximately 5 mm above its junction with the right subclavian vein.

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