CASE REPORT

The role of radiology in diagnosing synchronous subglottic and mediastinal haemangioma: a rare clinical entity

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Abstract

Subglottic haemangioma is a rare and life-threatening condition in infancy. A four-month-old girl presented with cough and wheezing with a history of three prior hospitalisations for bronchiolitis and inspiratory stridor. An upper airway endoscopy and imaging, including ultrasonography, CT scan, and MRI, revealed synchronous subglottis and mediastinal haemangioma. She responded to oral Propranolol treatment with complete resolution within six months of initiating therapy. Subglottic haemangiomas should be considered in infants with recurrent airway symptoms and obstruction.

Keywords: Subglottic haemangioma, Airway obstruction, Infant, Computed tomography, Magnetic resonance.

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Introduction

Infantile haemangioma is the most common head and neck tumour in infancy. Although present at birth, laryngeal haemangiomas usually become symptomatic within the first six months of the proliferative phase with airway obstruction and stridor. While half of the affected children have cutaneous haemangiomas, concurrent mediastinal haemangioma extending into the subglottic region is rare.[1]

Broncholaryngoscopy is the primary diagnostic tool for airway haemangiomas, which are typically seen as a blue-to-pink soft tissue mass protruding into the lumen.[1] However, submucosal or subglottic haemangiomas may not be detected with broncholaryngoscopy.[1] In such cases, radiological investigation can show the lesion and its extensions. CT provides high spatial resolution for airway assessment, while MRI better characterises soft tissue masses.[2]

This case report discusses the contribution of radiological modalities in diagnosing subglottic haemangioma with rare coexistence with mediastinal haemangioma.

Case report

A four-month-old girl, born at term with a birth weight of 3,200 grams, presented with persistent cough and wheezing. Initially, in April 2022, at two months of age, she presented at the Paediatric Emergency Unit of Faculty of Medicine at Ege University in İzmir. Broncholaryngoscopy was performed due to a preliminary diagnosis of laryngomalacia, but the findings were normal. She had been hospitalised three times for bronchiolitis.

She presented to İzmir Dr. Behçet Uz Children's Hospital for Paediatric Diseases and Surgery, in June 2022, at four months of age. On physical examination, she had coarse breathing sounds with inspiratory stridor. The oxygen saturation was 80% in room air by pulse oximetry. Inhaler treatment with a high-flow nasal cannula (HFNC) using a fraction of inspired oxygen (FiO2) of 30% and 2 l/kg/min was begun. Routine laboratory tests and chest X-rays were normal. Ultrasonography revealed a hypoechoic solid lesion encasing the left carotid artery without luminal obstruction. Contrast-enhanced CT showed a 6x8x11mm hyperdense subglottic lesion narrowing the airway and a 23x20x64mm hyperdense mass extending from the left carotid sheath to the anterior mediastinum. MRI confirmed the lesions as haemangiomas (Figure 1).

Oral Propranolol (2 mg/kg/day) was initiated, leading to rapid improvement of symptoms by day one. By day three, she was asymptomatic and oxygen therapy was discontinued. By the seventh day, ultrasonography showed significant reduction of the lesion (from 23x20x64mm to 8x7x17mm). She remained asymptomatic on follow-ups, with complete regression on CT at six-month follow-up, allowing discontinuation of Propranolol (Figure 1). Written consent for publication of this case report was obtained from the patient's parents.

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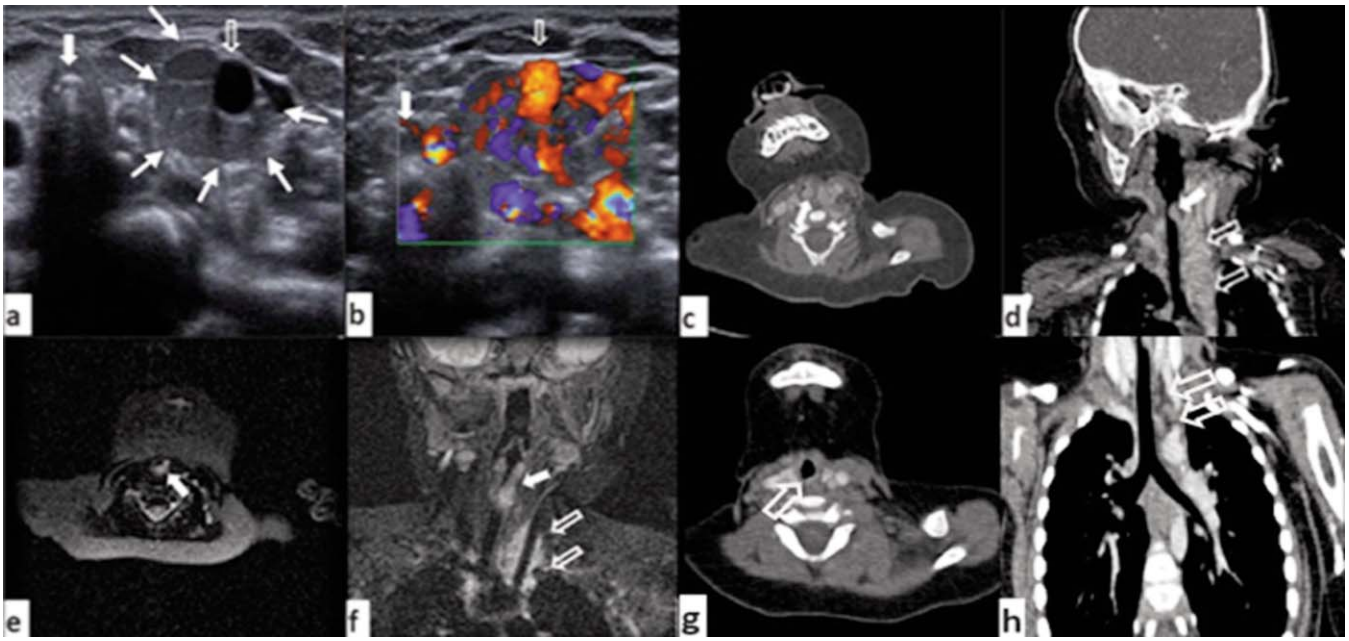

Figure-1: a) Grey-scale ultrasonography shows a soft tissue mass (arrows) surrounding the carotid sheath, encasing the left main carotid artery (empty arrow) and adjacent to the trachea (solid arrow). b) Colour Doppler reveals the lesion's hypervascular nature with arrows indicating the left main carotid artery and trachea. c) Contrast-enhanced CT shows an enhanced soft tissue mass (solid arrow), narrowing the upper airway. d) Coronal reformatted CT image shows a similar mass extending along the carotid sheath into the mediastinum (empty arrows) and the glottic mass (solid arrow). e) Axial T2-weighted MRI shows a hyperintense lesion (solid arrow) narrowing the airway. f) Coronal T2-weighted MRI shows a concomitant haemangioma (empty arrows) around the left main carotid artery extending to the mediastinum and the glottic mass (solid arrow). g) Axial and h) koronal post-treatment contrast-enhanced CT shows the disappearance of the hypodense mass (empty arrows).

Table-1: Case reports of concurrent subglottic haemangioma and mediastinal haemangiomas.

Feature	Case 1(Truong) (2010)	Case 2(Tamagna) (2011)	Case 3(Onder) (2019)	Case 4(Link) (2021)	Our case
Sex	Male	Female	Female	Female	Female
Age	One month	Six months	Two months	One month	Four months
Clinical findings	Feeding difficulties, dyspnoea	Reflux and recurrent respiratory infections	Cough and dyspnoea	Dyspnoea	Cough, dyspnea and recurrent respiratory infections
Imaging	MRI	CT	CT	MRI	CT and MRI
Radiological findings	A large intrathoracic haemangioma that narrows the airway covers the cervical midline structures and extends to the mediastinum.	A heterogeneous, highly vascularised mass in the left hemithorax.	A large mediastinal enhancing mass compresses the airway.	An intense vascular mass surrounds the great vessels in the right upper mediastinum. It compressed the trachea externally and narrowed the airway.	CT:Hyperdense subglottic lesion indenting airway.Soft tissue mass from left carotid sheath to the anterior mediastinum, showing contrast enhancement.MRI:Lesion extending from carotid bifurcation to mediastinum.Hypointense on T1, hyperintense on T2.
Endoscopy	A submucosal mass obstructed the subglottic airway by 80% and extended 2 cm below the trachea.	A large pulsatile mass that obstructed approximately 80% of the larynx and trachea	Shortened aryepiglottic folds and right-sided subglottic multiple haemangiomas	A vascular lesion with a compressing subglottic airway	Normal
Surgical treatment	Carbon dioxide laser ablation, subglottic resection	No surgery	Glottoplasty	No surgery	No surgery

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Medical treatment	Dexamethasone: Not therapeutic.Prednisolone and Propranolol: five-month combination therapy	Betamethasone: Not therapeutic.Switched to oral Propranolol	Oral Propranolol	Oral Propranolol administration for 6 months	Oral Propranolol
Follow-up	Symptoms relieved in two days.Mass decreased by 50% in 1 week.Symptoms disappeared at five-month follow-up.	Symptoms were relieved in five days and disappeared at one year of follow-up.	Symptoms were relieved in three days and disappeared at one year of follow-up.	Symptoms relieved in 3 days.Mass decreased by 50% in one month.Symptoms disappeared for six months.	Symptoms relieved in one day.Significant lesion reduction in one week (USG).Symptoms disappeared in six months.No mass was observed in six months CT.

Discussion

This case highlights the importance of radiological evaluation in infants with recurrent airway symptoms despite normal bronchoscopy. While bronchoscopy is useful, submucosal and deeply seated lesions may be missed, necessitating imaging for accurate diagnosis. The combination of USG, CT, and MRI played a key role in detecting synchronous subglottic and mediastinal haemangiomas.

An endoscopic examination can directly visualise the lesion and evaluate the airway, but it is invasive, difficult in neonates, and may miss submucosal or deep haemangiomas. Computed tomography was recommended as the primary diagnostic method in such cases.[3,4] Koplewitz et al. compared the performance of CT and endoscopy in the same paediatric patients and showed that CT detected all cases while 27% of cases were missed by bronchoscopy.[3] Choi et al. reported that CT may be helpful, especially in diagnosing submucosal airway haemangiomas undetected with laryngoscopy.[4] The current case was consistent with the literature. The baby had a normal laryngoscopy, but CT detected synchronous submucosal and mediastinal haemangiomas. Subglottic localisation or submucosal placement was the potential cause of being overlooked in endoscopy.

There are four reported cases in the literature of concurrent subglottic haemangioma and mediastinal haemangioma (Table I).[5–8] In two cases, the subglottic haemangiomas were detected primarily by endoscopy, followed by surgical procedures. One case underwent laser ablation and haemangioma resection, and one case underwent glottoplasty. When the symptoms did not improve, CT revealed synchronous mediastinal haemangioma.[7,8] In the other two cases, the diagnosis of both subglottic and mediastinal haemangiomas was initially made by CT, followed by a laryngoscopic examination.[5,6] All infants were treated with Propranolol.[5–8] The current case aligns with similar reports, reinforcing the effectiveness of Propranolol therapy and the necessity of radiological imaging for accurate diagnosis. CT is highly sensitive for assessing airway involvement, while MRI provides superior soft tissue contrast, aiding in differentiating haemangiomas from other masses.

Conclusion

Haemangiomas should be considered in infants presenting with persistent airway obstruction and recurrent infections. Imaging modalities, particularly CT and MRI, are indispensable for diagnosis, localisation, and treatment monitoring. Early Propranolol therapy results in favourable clinical outcomes, reducing the need for invasive interventions.

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YY, BDC & OB: Concept, design, data acquisition, analysis, interpretation, drafting, revision, final approval and agreement to be accountable for all aspects of the work.