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ABERRANT IN NOMINATE ARTERY A RARE CAUSE OF STRIDOR: CASE REPORT

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ABSTRACT

A boy 6 years old known case of cobalamin c disease admitted to ICU due to stridor and respiratory distress. ENT referral done to assess the upper airway and to rule out any cause of upper airway obstruction. Endoscopic examination was normal. CT scan chest with contrast ordered, and showing innominate artery compressing the trachea. This case reported to high light the possibility of this rare situation. We have to put this possibility in mind during the differential diagnosis of stridor causes especially if no clear cause is there.

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INTRODUCTION

Stridor is not a disease but it is a clinical symptom. It occurred when air flows through narrowed airways. Inspiratory stridor if the pathology on the glottis level and biphasic stridor when the pathology around sub glottis and trachea (Zawadzka-Głos, 2005). The narrowing of the air way may be due to pathology in the wall of respiratory tract or external compression of respiratory tract wall. Laryngomalacia is the most common cause of congenital stridor then followed by congenital vocal cord palsy and subglottic stenosis (Zawadzka-Głos *et al.,* 2005). Congenital vascular anomalies is very rare cause for stridor (Schuster *et al.,* 1991). Gross and Neuhauser first recognized an anomalous in nominate artery compressing the trachea and causing respiratory symptoms in 1948 (Gross and Neuhauser *et al.,* 1948).

CASE REPORT

6 years old boy whom is a known case of cobalamin c disease, suffering from recurrent attacks of convulsion, hydrocephalous, recurrent respiratory distress, chronic cough and recurrent chest infection. Patient referred to us to assess the airway because of stridor.

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Indirect laryngoscopy done and revealed free glottis and supraglottic areas. CT chest with contrast ordered and revealed the right brachiocephalic trunk emerges normally from the aortic arch but acquires an aberrant course (Fig 1). It turns medially just in front of the trachea at the sternum level. The anteroposterior diameter of the trachea is significantly reduced to almost 1/4th of its normal diameter (Fig 2&3). Patient then referred to vascular surgeon for further management.

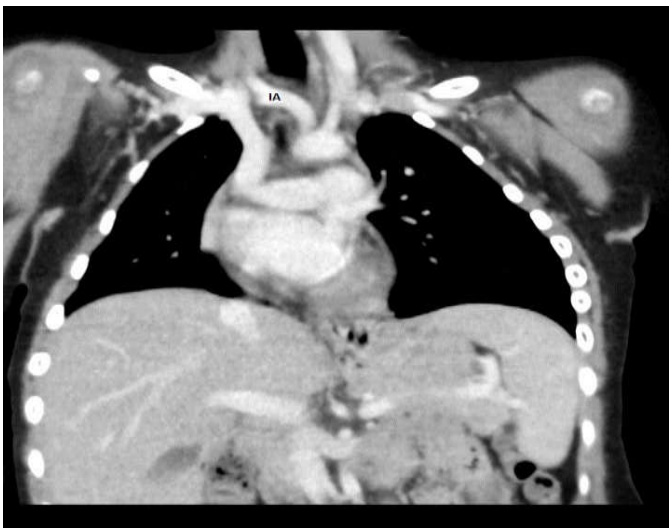

Fig 1. CT chest with contrast, coronal cut, showing innominate artery (IA) crossing the trachea

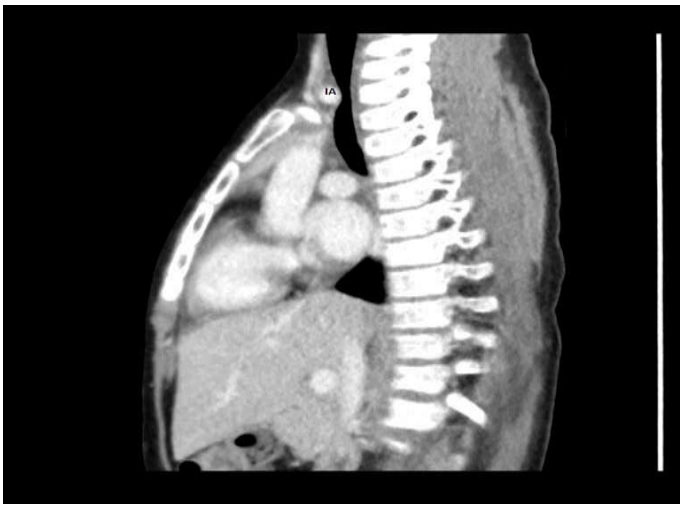

Fig. 2. CT chest with contrast, sagittal cut, showing innominate artery (IA) compressing the trachea

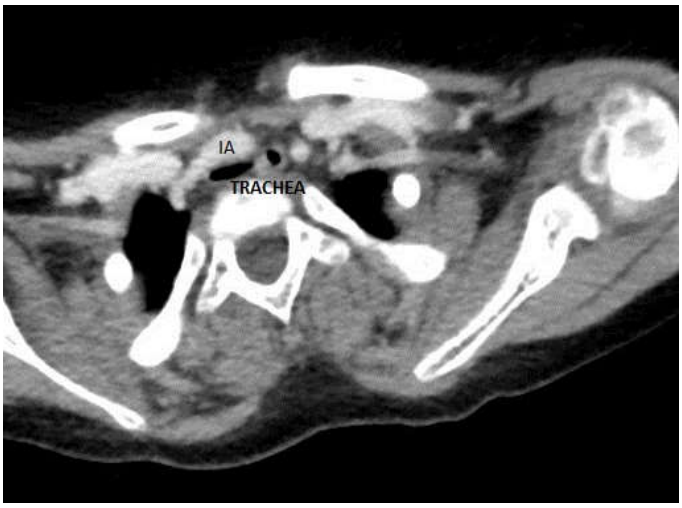

Fig. 3. CT chest with contrast, axial cut, showing innominate artery (IA) compression the trachea

DISCUSSION

Congenital vascular anomalies is very rare cause for stridor (Jabłońska-Jesionowska and Zawadzka-Głos, 2013). Gross and Neuhauser first recognized an anomalous innominate artery compressing the trachea and causing respiratory symptoms in 1948 (Gross and Neuhauser, 1948). Cough, stridor and occasionally apnea are the main symptoms (Gross and Neuhauser, 1948). Our case presented by stridor, chronic cough and recurrent chest infection. Schuster *et al.* 1991 reported airway compression by abnormal innominate artery as a cause of stridor (Schuster *et al.,* 1991). Our case the innominate artery was normal in origin but the course was aberrant. However, other authors believe that abnormalities in the tracheal wall is responsible for the symptoms (Fletcher and Cohn, 1989). The course of in nominate artery is aberrant in about 30% of normal children (Strife *et al.,* 1981). Mustard et al. reported only 13.6% of his patients with aberrant innominate artery need surgery because of marked symptoms (Mustard *et al.,* 1969). Diagnosis depends on various modalities like tracheoscopy, MRI and CT chest (Faust *et al.,* 2002). In our case, the diagnosis was established using CT chest with contrast.

Conclusion

Although it is a rare finding, but need to be in mind during examination of children with stridor because if you failed in the diagnosis this will cause delay in the management.

Conflict of interest: The authors declare that there is no conflict of interest regarding the publication of this paper.

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