

Clinical-Medical Image

A Rare Cause of Respiratory Distress in New borns: Laryngotracheobronchial Cartilage Calcification

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Abstract

The causes of respiratory distress are many in the new born; the malformative and infectious origins are the most frequently found, Laryngotracheobronchial cartilage calcification is a rare entity in infants. We report a case of a 2-month-old boy who presented to the paediatric emergency for respiratory distress, the clinical, morphological evaluation especially radiological findings identified a Calcifications of Laryngotracheobronchial tree, ear and larynx. Our purpose is to spotlight the importance of the appropriate clinical examination in new born because the origin is not always obvious.

Keywords: Laryngotracheobronchial cartilage calcification; Computed tomography; Chest radiography

Introduction

Laryngotracheobronchial cartilage calcification is a rare entity in new born, it is considered significant as it can cause respiratory difficulty and stridor especially in children under 1 year old. Abnormal calcification of cartilages in children is rare entity; it can be seen in Keutel syndrome, warfarin embryopathy, X-linked recessive chondrodysplasia punctate, or idiopathic. Keutel syndrome a rare autosomal recessive disorder, it is characterized by brachytelephalangism, abnormal cartilage calcification, and midfacial hypoplasia reported during imagery investigations outside the acute episode in our patient.

Case Report

We report a case of an 2-month-old boy who presented to the paediatric emergency for respiratory distress without fever, clinical examination finds an inspiratory and expiratory stridor and wheezing over both lungs on chest auscultation, associated with an the atypical craniofacial appearance characterized by midfacial hypoplasia with a broad depressed nasal bridge. A radiological assessment was carried out made of chest radiography (Figure 1) and shows the presence of tracheobronchial cartilage calcification without tracheobronchial stenosis confirmed by a thoracic computed tomography.



Figure 1: Tracheobronchial cartilage calcification on posteroanterior chest radiography.

A chest angiotomography was performed showed laryngotracheobronchial calcification (Figures 2 and 3) and stenosis of the right pulmonary artery (Figure 4), but no cardiac malformation or chest abnormality, a bronchoscopy was performed in the new-born later in the hospital, which confirmed the absence of tracheal lumen stenosis.

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Figure 2: Laryngeal calcification on axial CT section.

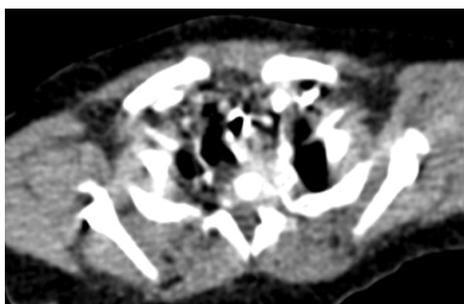


Figure 3: Tracheal calcification on axial CT section.



Figure 4: Decrease in the size of the right pulmonary artery (yellow arrow), and bilateral bronchial calcification.

A Braine and cervical tomography showed the presence of calcification laryngeal cartilages (Figure 4), but no calcification intracranial. Skeletal and limb X-ray showed cranium thickness, and discreet shortening of the distal phalanges of hands (Figure 5). The parents were phenotypically normal first cousins. The final assessment was laryngotracheal calcifications in a child with suspected Keutel syndrome. Genetic studies for confirmation of diagnosis have not been attempted as this facility is not available in our country.

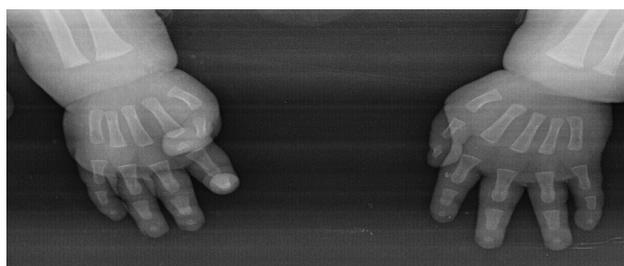


Figure 5: Shortening of the distal phalanges of the hands.

Discussion

Calcification of laryngotracheobronchial cartilage may develop due to degeneration of cartilage in asymptomatic elderly people and usually has no clinical significant, the reason for the premature calcification in children is not known especially in small infants under 02 Years old [1]. Some authors have reported that chronic respiratory tract irritation due to recurrent lung infections and genetic factors plays a role in the aetiology in children [1].

However, laryngeal calcification is considered significant as it can cause respiratory difficulty and stridor especially in children under 1 year old [1,2]. Some cases were reported related to congenital cardiovascular disorders, Keutel syndrome, chondroplasia punctata, warfarin embryopathy, and warfarin sodium therapy subsequent to mitral valve replacement [3].

Keutel syndrome characterized by abnormal cartilage calcification, brachytelephalangism, hearing loss, pulmonary stenosis, diffuse calcification

of the epiglottis, nose, larynx, tracheobronchial tree and cerebrum, and characteristic facial appearance [4], it is a rare autosomal recessive cartilage tissue disorder which described firstly in two siblings of consanguineous parents by Keutel in 1971.

Actually, it been demonstrated that MGP (Matrix γ -carboxyglutamate protein) gene mutations are responsible for Keutel syndrome by abnormal calcification of different cartilage areas. Matrix γ -carboxyglutamate protein (MGP) is a mineral binding and vitamin K depended ECM protein, and acts as a local inhibitor of calcification.

According to the literature data, the most common clinical manifestations of Keutel syndrome were cartilage calcification (100% of the cases), brachytelephalangism (100%), and facial dysmorphisms such as midrace retraction, depressed nasal bridge (94%), our patient also presented with abnormal cartilage calcification, facial dysmorphisms, and discreet brachytelephalangism. Recurrent upper and lower respiratory tract diseases, chronic sinusitis, chronic obstructive pulmonary disease, asthma, and Emphysema, are the main complications.

In summary, those clinical and imaging findings are extremely rare in a new born with respiratory distress. Imaging is important as it can demonstrate airway complications allowing for earlier intervention in an attempt to improve the patient's outcome.

Conclusion

The Follow-up in young children must include physical examination (facial appearance...) and complementary exams, especially assessment of upper and lower airway cartilage ossification, hearing and pulmonary function.

Declaration of Interests

The authors declare that they have no competing interests.

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