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Not to Miss a Subglottic Hemangioma in an Infant: A Case Report

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Abstract

Background: Subglottic hemangioma, especially in the absence of an evident skin lesion is a very rare congenital abnormality. Beside this rarity, the diagnosis is very challenging and needs assessment with rigid bronchoscopy and Computed Tomography (CT) angiography.

Case presentation: Here, we report the case of a 3-month-old boy with subglottic hemangioma, which was diagnosed after several visits and through several imaging procedures, including CT angiography and bronchoscopy visualization. Our case responded well to the medical treatment and was followed to find further regressions of the tumor.

Conclusion: When an infant presents with biphasic stridor and barking coughs within the first few months of the child birth, the diagnosis of a subglottic hemangioma should be kept in mind along with other differentials.

Key Words: Case Report, Male Infant, Subglottic.

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1- INTRODUCTION

It is reported that infantile hemangioma is the most common benign tumor present during infancy, affecting 4 to 5 percent of the total population (1). Although it is reported that head and neck regions are the most involved parts, the involvement of the subglottic region is very rare and constitutes only 1.5 percent of all congenital laryngeal abnormalities (2). It is also proposed that only 1 percent of the infants with cutaneous hemangioma may have subglottic hemangioma. However, one out of every two subglottic hemangioma cases, may have no cutaneous lesion (3).

This congenital malformation does not manifest itself immediately after the birth. Still due to its rapid growth, the clinical signs and symptoms develop within a few months after the birth. Noisy breathing and even biphasic stridor can be a non-specific symptom of the patients. Due to the high growth rate, subglottic hemangioma can even lead to a life-threatening condition. With this regard, the diagnosis is challenging and even may be time consuming. But it is important to make a timely diagnosis and avoid development of airway obstruction (4, 5).

Only a high suspicion and visualization of the typical feature of the tumor during bronchoscopy can lower the risk of missed diagnosis. However, the symptoms of the tumor can provide a vast number of differential diagnoses and many cases reach a final diagnosis after different clinical and paraclinical assessments (6). Here, we present a case of subglottic hemangioma that can be considered as a missed diagnosis.

2- CASE PRESENTATION

A 3-month-old Iraqi boy, born through cesarean section, presented with a chief complaint of noisy breathing starting three days after the birth. His mother visited a pediatrician with this regard and an outpatient prescription of saline drop was advised with a possible diagnosis of viral infections. However, the noisy breathing continued until the age of 2 and half months. Her mother stated that attacks of breathing shortness happened and the noises were changing according to the patient's position. Even another consultation with another pediatrician yielded to a misdiagnosis of allergic disease. The physician had prescribed salbutamol and augmentin syrups; however, no remission in the symptoms was reported after two weeks.

A third pediatrician was consulted in Iraq and the physician suspected laryngomalacia. So, the doctor prescribed Dexon and Allermine syrups and advised to visit an Eye, Nose, Throat (ENT) specialist for further workups. The medical treatment was partially effective. The patient underwent Neck and Chest magnetic resonance imaging (MRI) in order to exclude congenital anomalies of the respiratory tract. However, the MRI showed only a subglottic stenosis. The patient also underwent neck and chest computed tomography that was suspicious for tracheal stricture and the radiologist suggested a bronchoscopy for final diagnosis.

The patient traveled to Iran for further workups. He was visited by a thoracic surgeon and a rigid bronchoscopy showed tracheal stenosis just above the carina, around 1.25 cm below the vocal cord (Fig. 1). According to the lesion appearance, the surgeon decided to do CT angiography in order to exclude vascular anomalies. However, the CT angiography confirmed the presence of subglottic hemangioma. Treatment with corticosteroid and propranolol was started for the patient and after a monitoring of 48 hours no drug side effect appeared and the patient experienced a major relief from the symptoms. Therefore, further surgical therapies including laser resection were

considered as alternative therapies, if the medical treatment was not well responded.

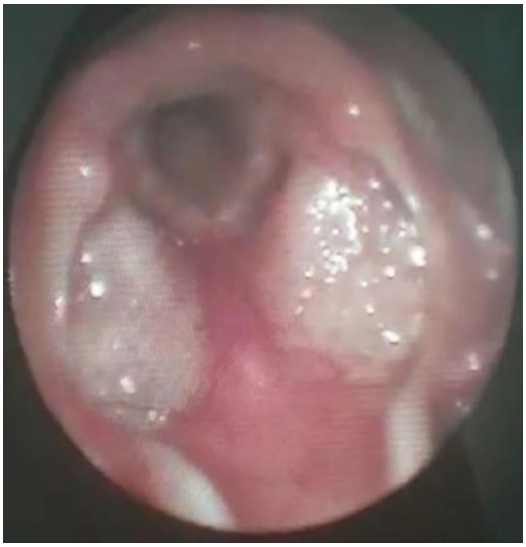

Fig. 1: Bronchoscopy feature of the lesion

3- DISCUSSION

The first report of subglottic hemangioma comes from 1913, when Phillips and Ruh (7) reported a case of subglottic hemangioma in a nine-month-old male. In fact, the condition may be diagnosed a few weeks or even a few months after the child's birth. The peak growth rate happens at the age of 1 month to 3 months and usually the growth is halted at the age of 5 months. The mean age of subglottic hemangioma diagnosis is 4 months. However, Lin et al. (6) reviewed 3 reports along their case and concluded that the age of diagnosis is ≤6 months. The tumor experiences regression within the few first years of the patient's life. Infantile hemangiomas are higher in female gender and Caucasian ethnicity. Other risk factors include prematurity, high maternal age, low birth weight, placental abnormalities, and preeclampsia (8, 9). Our case was a three-month-old boy and was diagnosed at the age of 3 months.

It is reported that patients with beard distribution of hemangioma or those who have anterior neck and oral and/or pharyngeal mucosal hemangiomas are at higher risks of developing upper airway lesions (10). However, half of the patients with subglottic hemangioma have no cutaneous manifest. Moreover, the manifested signs and symptoms are also non-specific. Infants are, often, presented with difficulty in breathing, wheezing, stridor, and even cyanosis (6). Bizarre presentations like difficulty in feeding are also reported in the literature (11). With this regard, in the presence of croup-like illness without any cutaneous involvement, subglottic hemangioma should be suspected (1, 9). The case in our study was also presented with biphasic stridor, and a list of differential diagnoses was ruled out through assessments of different physicians and even in a multinational manner to reach the final diagnosis of subglottic hemangioma.

This bizarre demonstration of the disorder causes delayed diagnosis and an imposed rate of morbidity and mortality. The reported rehospitalization and death rates were reported to be 30 and 2 percent, respectively (6, 12). Darrow et al. (2) tried to propose an algorithm for the management of the subglottic hemangioma. They reported that in the absence of the beard distribution hemangioma in the face and biphasic stridor or barky cough, there is no need for further evaluation. However, even in the presence of only non-healing biphasic stridor and barky cough, without cutaneous lesion, further studies with awake flexible bronchoscopy should be done. During the rigid bronchoscopy, a reddish-blue color lesion may be detected; however, the lesion may be missed, especially when the child is on corticosteroid therapy or has an endotracheal tube. Thus, a CT angiography can be a complimentary helpful modality.

When the diagnosis is made, the treatment should be initiated. Corticosteroids along with propranolol are the routinely used

medical therapies. The prescribed dose of propranolol is 2–3 mg/kg/day divided into 2 daily doses maintained for at least 6 months (13, 14). Moreover, it is advised to continue the treatment until the age of 10 to 12 months and then wean the treatment and follow the patient for possible rebound growth (1, 15). However, if the medical treatment does not respond well, a surgical resection including open or laser resection should be planned. Also, it is reported that nonselective beta-adrenergic receptor antagonists are well tolerated in these patients and side effects like hypoglycemia, bradycardia, hypotension, and biochemistry test changes barely happen among the patients (9, 16). The patient in our report responded well to the treatment with no major adverse effect.

4- CONCLUSION

When an infant presents with biphasic stridor and barking coughs within the first few months of the child birth, the diagnosis of a subglottic hemangioma should be kept in mind along with other differentials. Our case was finally diagnosed through rigid bronchoscopy and CT angiography. The response to medical treatment was good in the patient; but, in case of failure during the future follow up, surgery should be chosen alternatively.

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