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Mediastinal Lobular Capillary Haemangioma in Children with Severe Airway Obstruction Symptom: A Case Report

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ABSTRACT

Background: Lobular capillary haemangioma (LCH) is a polypoid form of capillary haemangioma occurring on the skin and mucosal surfaces. While LCH of the oral and nasal cavity is well-known entity, mediastinal localization is a rare case. In children, LCH is frequently benign.
Case illustration: A two month age baby girl experienced recurrent inspiratory stridor since she was 42 days-old and diagnosed as laryngomalacia. Due to the widening of mediastinal area from chest x-ray, a chest CT-scan was conducted and showed a mass in the mediastinal area. A sternotomy was done and found mass filled the mediastinal space. Some parts of the mass found to depress the trachea. Mediastinal tumor resection procedure done. A definitive pathology anatomy result was a lobular capillary haemangioma. After surgery, stridor gradually improves.
Summary: We reported this rare mediastinal lobular capillary haemangioma case that was initially diagnosed with laryngomalacia After removal of the tumor, the stridor gradually improved.

Keywords: Lobular capillary haemangioma, pyogenic granuloma, mediastinal tumor, stridor, children

INTRODUCTION

Patient with stridor can be caused by number of differential diagnoses ranging from mild to severe pathology.[1] Mediastinal masses include a wide variety of tumors occurring in patients of all ages that have clinical presentation of mediastinal masses can range from being asymptomatic to symptoms due to compression or direct invasion of surrounding structures. The main symptoms associated with the mediastinal tumors include cough, chest pain, and dyspnea.[2] One kind of the mediastinal tumors is a lobular capillary haemangioma (LCH). LCH is a polypoid form of capillary haemangioma occurring on the skin and mucosal surfaces, LCH also known as pyogenic granuloma, it's kind of benign, typically painless tumors.[3] Hemangiomas account for approximately 0.5% of all mediastinal tumors.[4] Radiological characteristics could be an early important

component for suggestive diagnosis. Definitive diagnosis relies on histopathology examination, which reveals variably sized capillaries proliferation. Surgical treatment is usually required because this lesion rarely resolves spontaneously or can give severe symptom.[1] In this case, we present rare case of lobular capillary haemangioma who experienced recurrent inspiratory stridor. After removal of the tumor, the stridor gradually improved.

CASE ILLUSTRATION

A two month age girl referred to emergency department due to recurrent inspiratory stridor since she was 42 days-old. Patient was initially complained for dyspnoea with stridor and chest retraction. Patient also complained about dry cough. There are no bluish appearance while dyspnoea. There are no history of fever. This complained is the second episode of stridor symptom. Patient already got previous episode of stridor and said improve after got hospitalization. At first episode, patient was hospitalize for 9 days, patient discharge after improvement of symptom. Just 4 days later, the symptom was reappear, and after 3 days of hospitalization, patient referred our Hospital. Patient was alert with Respiratory rate 52 times per minutes and pulse rate 172 times per minutes. Stridor was heard in inspirational phase. There was retraction on subcostal, intercostal, and suprasternal part of the chest. From X-ray examination there was suspected a mass at anterior mediastinum showed by widening of the mediastinal area (Figure 1). Multislice computed tomography (MSCT) with contrast (Figure 2) was done that showed soft tissue mass with indeterminate border at regio right and left colli (dominant left) that expand to the anterior mediastinum strengthening inhomogeneity after contrast injection, and mass cause compression of the larynx and pushes the trachea to the right, concluded with suspicion of fibromatosis colli differential diagnose with neurofibroma and lymphoma. Laboratory examination revealed white blood cells 20.07 x 10^3/µL, neutrophils 18.41 x 10^3/µL (91.74%), lymphocytes 1.18 x 10^3/µL (5.86%), haemoglobin 10.71 g/dL, MCV 91.35 fL, MCH 27.81 pg, MCHC 30.44 g/dL, haematocrit 35.19%, RDW 13.09%, platelet 413.5 x 10^3/µL. Blood gas analysis revealed pH 7.39, pCO2 58.1 mmHg, pO2 185.60 mmHg, BEecf 9.1 mmol/L, HCO3- 34.1 mmol/L, SO2c 99.2%, TCO2 35.90. Electrolyte serum revealed Natrium 138 mmol/L, Kalium 4.94 mmol/L, Chlorida 96.3 mmol/L, Calcium 10.1 mmol/L. SGOT 31.8 U/L, SGPT 32.3 U/L. BUN 6.20 mg/dL, creatinine 0.27 mg/dL. Serology Beta HCG <0.10 mIU/mL and Alfa feto protein 1042 IU/mL.

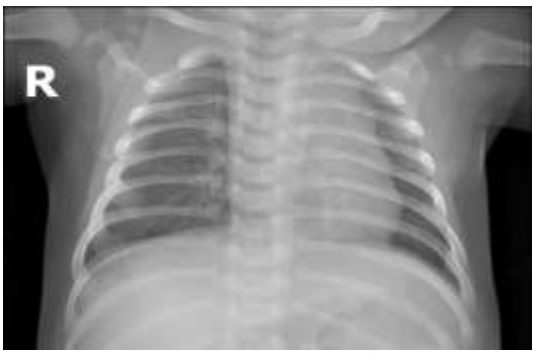


Figure 1. Chest X-ray shows widening of upper mediastinum space.

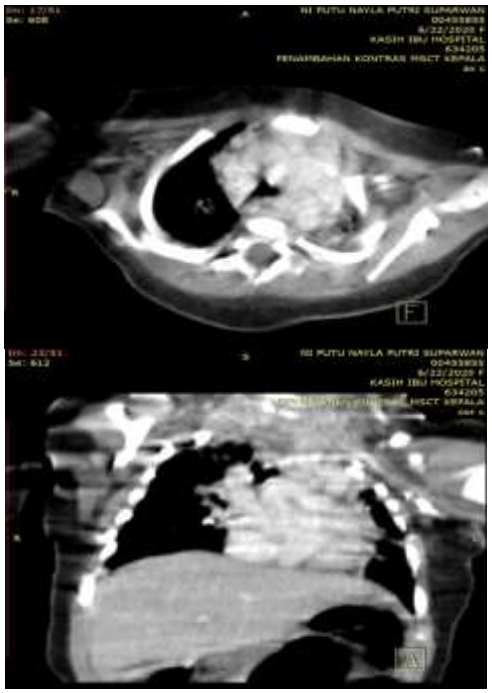


Figure 2. Thorax CT scan revealed soft tissue mass on Regio coli (dominant on the left side) which extend to anterior mediastinum and push the trachea to the right side.

At 2nd day hospitalization patient underwent sternotomy and mediastinal tumor resection procedure. The tumor tissue was taken for histopathological examination (Figure 3). The procedure last without significant surgical complications. After procedure patient still intubated and continue treatment at PICU. Additional antibiotics and analgetic therapy were given. Histopathological examination (Figure 4) revealed proliferation of small blood vessel with various sizes forming multinodular pattern that lined by fibrous connective tissue septa. That blood vessel is lined with endothelial cells with a round, ovoid-spindle morphology that tend to be bland, eosinophilic cytoplasm. There was also mature fatty tissue in the vicinity and it was concluded a vascular lesion tend to be capillary hemangioma.

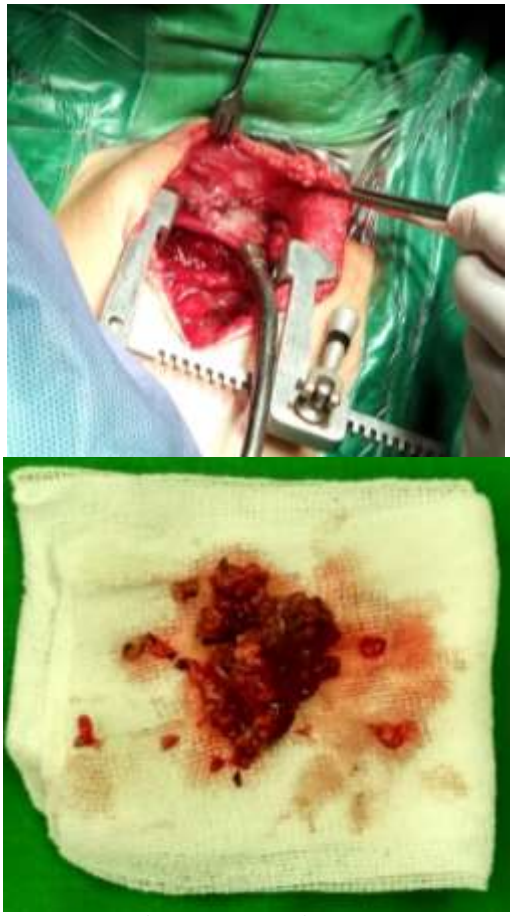
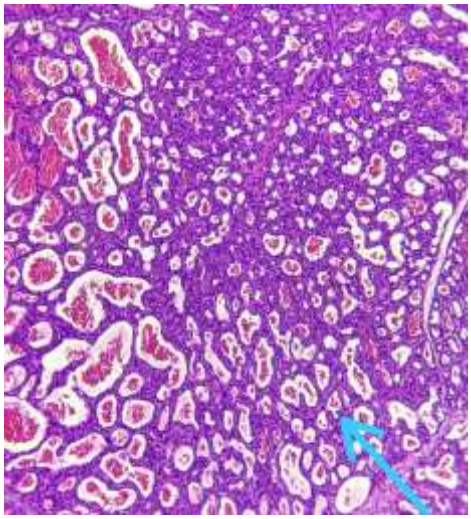


Figure 3. Mass of haemangioma after completely resected.



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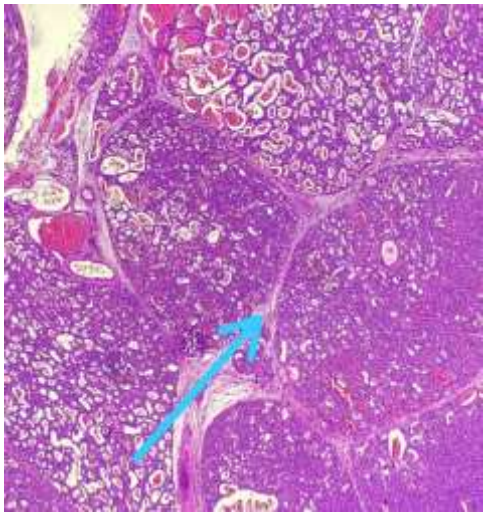

B

Figure 4. A. Proliferation of small blood vessel. B. Multilobular pattern with fibrous connective tissue septae.

At 3rd day hospitalization, patient got improvement of the clinical symptom and decided to stop steroid injection. The other therapy continued. At day 6th hospitalization, patient been extubated after good response from weaning of ventilator setting. Then the oxygen therapy was switched to simple mask.

DISCUSSION

Patient with mediastinal tumors can has asymptomatic problem or symptom with range from mild to severe feature causes by compression or direct invasion of surrounding structures. The main symptoms associated with the mediastinal tumors include cough, chest pain, and dyspnoea.

Mediastinal tumors can represent a wide and various histological group include neoplastic or benign lesion. One kind of the mediastinal tumors is a lobular capillary haemangioma (LCH). LCH account for approximately 0.5% of all mediastinal tumors. LCH is a benign vascular tumors from the skin or mucous membranes characterized by rapid growth and friable surface. [1,2,4,5] Some study showed LCH mostly seen in female with ratio male to female for LCH was 1:1,2.[6,7]

The etiology of this tumors is remain unclear. Although the name suggests an infectious etiology, the cause of pyogenic granuloma (PG) is unknown. Some report state that includes trauma, drug side effects, hormonal, viral oncogenes, production of angiogenic factors, abnormalities of cytogenetics, and local irritation. This study also stated about production of angiogenic vascular growth factor such as vascular endothelial growth factor (VEGF) signal transduction pathway proteins are overexpressed as pathogenesis of LCH. But the exact role of the growth factor still undetermined.[6,8] One study that analyse 11 patient with LCH revealed embryonic stem cell markes in the endothelial cells and more differentiated pattern in the interstitial cells, suggesting de novo vasculogenesis from the primitive stem cells.[9] Pagliai report about investigation the clinical characteristics and natural history of LCH. The report showed that around 76.9% lesion found on the head and neck area, 76,8% had no history of trauma or other predisposing factor, and mostly manifested as a single lesion.[10]

Recurrence inspirational stridor was main complaint of this patient that needs to referred to our hospital. Stridor is an abnormal, high-pitched monophonic sound, produced by turbulent airflow caused by the oscillation of a narrowed airway. Its presence suggests significant obstruction of the large airways at the level of the supraglottic, glottis, subglottic, or trachea. Inspiratory stridor is one kind of stridor that originating from obstruction in the extra thoracic region. Stridor is observed with subglottic localization of the tumor.[11]

This kind of mediastinal tumor can occur at some of our body part like skin surface and mucosal membrane. But mostly this lesion located at the skin. One report about 639 case of vascular lesion located in the mouth, upper respiratory tract, throat, and trachea, that found 73 cases of LCH. LCH lesion are very rare located in the airway, with only 11 cases reported in the trachea and left main bronchus.[8] Imaging studies frequently used to evaluate the differential diagnosis patient with dyspnoea. Some of the picture with the widening of the mediastinal area at chest x-ray. CT imaging is the modality of choice in identifying a mediastinal masses. This modality could define seat, size, density, and relationship with other anatomical at chest. It is also as a guide for further biopsy or excision procedure. LCH has a the non-specific features of a well-defined soft tissue density mass with a hypoattenuating cap of variable thickness.[7,12] A homogeneously marked enhancement is observed after contrast injection.[3]

Histopathological examination of the mass is recommended.[13] Histopathological examination of LCH mainly find numerous lobular capillary proliferation in an oedematous fibroblastic stoma along with inflammatory cells.[14,15] Although spontaneous regression may occur in some patients, treatment is usually required for LCH because as the consideration of bleeding or symptom severity on the patient. There is no consensus regarding the optimal approach.[16] Study from Lee revealed that from 753 patient with LCH were treated surgically with majority of the case was managed by surgical excision.[17] Sternotomy is recommended for anterior mediastinal tumor excision because it provides enhanced exposure, which indirectly improves safety and pain control and is not associated with long-term thoracic deformity.[18]

CONCLUSION

Patient with stridor can be cause by number of differential diagnoses ranging from mild to severe pathology. Important to make some differential diagnosis and consider proper further examination. Mediastinal tumor should be included in the vast differential diagnosis when evaluating stridorous children. LCH is one kind of mediastinal tumors form of capillary haemangioma occurring on the skin and mucosal surfaces. The choice of therapy should consider the clinical condition of the patient whether there was a risk of complications such as respiratory complications.

Declaration by Authors

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