Acta Scientific PAEDIATRICS (ISSN: 2581-883X)

Volume 6 Issue 6 June 2023

Case Report

Clinical Case of Tracheal Leiomyoma in A 14-Year-Old Teenager

Evgeny Furman*, Ekaterina Khuzina, Natalya Grymova and Mariya Ponomareva

Federal State Budgetary Educational Institution of Higher Education "Academician Ye.A. Vagner Perm State Medical University of the Ministry of Healthcare of the Russian Federation", Perm, Russia

***Corresponding Author:** Evgeny Furman, Federal State Budgetary Educational Institution of Higher Education "Academician Ye.A. Vagner Perm State Medical University of the Ministry of Healthcare of the Russian Federation", Perm, Russia.

Received: March 22, 2023

Published: May 24, 2023

Abstract

Grounding: Primary tumors of the trachea and main-stem bronchi are rare and comprise from 1 to 2% of all respiratory tract tumors. In adults, the majority (60-90%) of these tumors are malignant, while in children, benign neoplasms are more common. About 1% of the primary tracheal tumors is leiomyoma. This neoplasm can cause obstruction of the upper respiratory tract and fatal asphyxiation.

Description of Clinical Case: A rare clinical case of tracheal leiomyoma in a 14-year-old teenager admitted to the Department of Pulmonology of the Regional Children's Clinical Hospital of Perm with complaints of shortness of breath is presented.

Conclusion: The case presented demonstrates a rare cause of obstruction of the upper and lower airways - tracheal leiomyoma, which requires timely diagnosis and special treatment tactics.

Keywords: Clinical Case; Obstruction; Trachea; Tumor; Leiomyoma; Asphyxia; Stenosing Laryngotracheitis

Grounding

Different diseases and damages can be the causes of airway (upper and lower) obstruction. Airway obstruction can lead to asphyxia and lethal outcome. In those cases, when air flow obstruction while breathing occurs in the oral, pharyngeal or laryngeal cavities, respiratory disorders are considered in context of the upper airway obstruction. The causes of the upper airway obstruction (Rogers M.C., 1995) in children may be the following:

- Congenital diseases:
    - Narrowing of internal airway lumen: subglottic stenosis, membrane, cyst, laryngocele, tumor, laryngomalacia, laryngotracheoesophageal membrane, tracheomalacia, tracheoesophageal fistula;
    - External squeezing and damages: vascular ring, cystohygroma;
    - Birth injury;
    - Neurological disorders;
    - Craniofacial anomalies;
    - Hypocalciemia.
- Acquired diseases and damages:
    - Infections: stenosing laryngotracheitis, epiglottitis, retropharyngeal abscess, Ludwig's angina, mycotic infection, paratonsillar abscess, diphtheria, bacterial tracheitis;
    - Injury: postintubation edema, posttracheostomic stenosis;
    - Burns of airways (thermal or chemical);
    - Aspiration of foreign bodies; • systemic disturbances; • tumors;
    - Neurological damages;
    - Chronic upper airway obstruction;
    - Hyperthrophic tonsillitis and adenoids [1].

Primary tumors of the trachea and main-stem bronchi are rare and make from 1to 2% of all respiratory tract tumors. In adults, the majority (60-90%) of these tumors are malignant, while in children, benign neoplasms are more common [2]. About 1% of the primary tracheal tumors is leiomyoma [3]. In contrast with esophageal leiomyoma, which is rather easy to diagnose, tracheal leiomyoma is a very rare disease, especially in children. Tracheal leiomyomas occur from the smooth muscle cells of the organ wall and more often are located in its lower third. Most commonly, they occur in men after 30 (all in all, less than 30 cases in adults were described) [4].

Symptomatology of tumors, as a rule, is connected with obstruction of the upper airways (dyspnea, more often of inspiratory type, acute respiratory failure, rales, stridor, relapsing pneumonia etc.). Nonspecific symptoms, are commonly cough, rarely - hemoptysis. Clinically, this symptomatology can mistakenly predict the diagnosis of bronchial asthma or chronic obstructive pulmonary disease (COPD). In rare cases, tumors of trachea and main-stem bronchi can be asymptomatic and detected accidentally [5-8].

We have not found out any cases of child leiomyoma, described in available Russian and English literature. The clinical case of the upper airway obstruction developed as a result of tracheal leiomyoma is presented below.

Clinical example

A boy D. born in 2004 was admitted to the Department of Pulmonology of the Regional Children's Clinical Hospital of Perm in September 2018 with complaints of noisy breathing and distant rales. The disease anamnesis: for the first time, the episodes of noisy breathing occurred in January 2018; by April 2018, attacks of heavy breathing with distant rales occurred during physical exercise, but the boy felt well. In April 2018, parents addressed pediatrician for the first time, the child was sent to allergologist-immunologist for consultation. During objective examination, disseminated dry rales were heard; on the basis of questioning and anamnesis data moderate bronchial asthma was diagnosed.

Since April 2018, the child received combined preparation (formoterol+budesonide) without any dynamics; attacks of dyspnea and dry cough occurred with minor physical exercise and at night. Since June 2018, montelukast was added to combined inhalation corticosteroids; intensification of basic therapy did not promote significant clinical improvement. In September 2018, after falling ill with respiratory infection, there was observed a negative dynamics in child's health status - dry cough, attacks of heavy breathing at rest, constant distant rales, which were not connected with the position of the body and physical exercise.

From patient's life history: child was born from the third pregnancy against the background of arterial hypertension, the third term labor. The birth weight was 6355 g, height 58 cm. The Aprag score - 7/8. At the age of 1 year and 1 year 2 months, he had obstructive bronchitis, at the age of 2 years and 1 month - pneumonia in the upper lobe to the left; at the age of 2 years and 6 months - acute laryngotracheitis with degree 1 laryngeal stenosis. Allergic anamnesis: calm. Heredity: not burdened.

When the boy was admitted to the department of pulmonology in 29.09.2018, his health status was moderately severe. He took forced position while eating - straightened his neck and slightly threw his head back. Thorax was of emphizematous form; neck and sternocleidomastoid muscles participated in breathing. Voice was loud. Distant inspiratory rales. Respiratory rate - 24 per minute. SpO_2 98%. Nasal breathing was heavy. Percussion sound above the lungs had vesiculotympanic resonance. Auscultation results: respiration - heavy, goes to all departments; expiration long, disseminated dry rales from all the sides. Heart rate: 76 per minute. Heart sounds: muted, rhythmic.

At the department of pulmonology, the following examination was carried out: complete blood test: erythrocytes $5.29 \cdot 10^{12}$/l, hemoglobin 146 g/l, leukocytes $11.6 \cdot 10^{9}$/l, eosinophils 1%, stab neutrophils 2%, segmented 65%, lymphocytes 24%, monocytes 8%, thrombocytes $321 \cdot 10^{9}$/l, ESR 5 mm/h; complete urine test: no pathology. In biochemical blood analysis, elevated level of glucose - to 6.6 mmol/l, the rest indices are within the normal values. Spirography: FVC 100%, FEV1 106%, PEFR 25-50-75 71-101-142%, broncholytic test - negative. Blood immunoglobulin test: elevated total IgE-522.0 ME/ml, elevated level of specific IgE to hen's egg and house dust ticks. Rhinocytogram: eosinophils 5%. Roentgenology: no infiltration.

Treatment received at pulmonological department: ipratropium bromide and fenoterol inhalation solution - 15 drops 4 times a day using nebulaser, beclometasone inhalation - 400 mg twice a day with nebulaser, mometasone - 100 mcg into each nasal passage once a day, loratadine - 10 mg once a day perorally, two intramuscular injection of dexamethasone in the dose of 0.5 mg/kg. Against the background of the therapy implemented, there was no dynamics in patient's health status - bronchoobstructive syndrome was clinically arrested, however, distant inspiratory rales at rest preserved, saturation was kept within 97-98%.

05.10.18. Diagnostic bronchoscopy. Tracheal aperture was free. Vocal cords were not changed; there was a neoplasm 2.5 cm distant from the cords, which closed the tracheal lumen by 2/3; it was of soft-elastic consistency, pink-white color, with marked vasculature, measuring up to 1.5 cm. The neoplasm was located on the posterior wall of the trachea.

05.10.18. Computed tomography of the chest of thoracic organs and MRT of the neck and trachea.

Conclusion. Endotracheal large neoplasm from the posterior wall of trachea at the level of vertebrae C6-C7 (Figure 1).

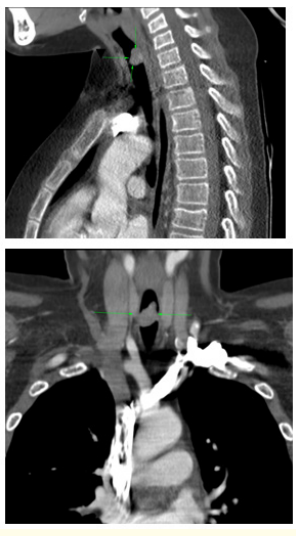

Figure 1: Patient's tracheal leiomyoma on the sagittal and frontal section by CT data.

The child was urgently transferred to surgical department.

05.10.2018. In connection with a high risk of asphyxia, tracheostome was applied, after the tracheostomic tube was installed, dyspnea was arrested. Biopsy, recommended by pediatric oncologist, was rejected for a time in connection with vascularization of neoplasm and threat of hemorrhage and respiratory disturbances. Blood α-fetoprotein, CGT and neuron-specific enolase was tested - the results were within normal values. At the surgical department, the boy received antibacterial and symptomatic therapy.

17.10.18. For further treatment, the boy was transferred to the Department of Thoracic Surgery of the City Children's Clinical Hospital №13 named after N.F. Filatov, Moscow. There was implemented a computed tomography of the thoracic cavity with intravenous contrast: the lumen in the upper third at the level of thyroid gland was totally narrowed at the expense of hypervascularized soft-tissue neoplasm measuring 18x14x9.6 mm without reliable signs of extension beyond the trachea; no changes and hyperplasia in intrathoracic lymph nodes were noted; a single axillary lymph node to the right was enlarged to 11-12 cm without any signs of pathological contrast. Repeatedly implemented bronchoscopy: no dynamics vs 05.10. 2018 It was decided to perform endoscopic excision of neoplasm using laser coagulation. 23.10.18. Surgery. 24.10.18. Decannulation.

The postoperative period was smooth; the patient received antibacterial therapy. On the day 6th after the surgery, the boy was discharged from the hospital in satisfactory state. Histological study of the neoplasm fragment was conducted at "Children's City Clinical Hospital №13 named after N.F. Filatov". The conclusion received was a spindle-cell tumor with uncertain histogenesis. For the final diagnosis, it was recommended to implement immunohistochemical study on the basis of Moscow Children's City Clinical Hospital.

04.12.18. The result of immonohistochemical study: tumorous tissue of medium cellularity with multiple calcified foci and moderate inflammatory infiltration; the tumor was partially covered with respiratory epithelium. The cells have light ovoid and spindle-shaped nuclei and prolate bipolar cytoplasm. The cellular elements of the tumor form fascicles. Mitotic activity was not determined. Diffuse expression by neoplastic cells SMA, desmin, calponin was received. The elements of macrophage line of the inflammatory background expressed CD68, factor XIIIa. The tumorous cells were negative in reactions with antibodies to ALK, CD34, melan-A, S100. Proliferative index Ki-67 was less than 5%. The final histological diagnosis: histopathological feature and immune phenotype confirm leiomyoma.

Discussion

The presented clinical example of a rare case of tracheal leiomyoma in a 14-year-old teenager demonstrated a complicated differential diagnosis of heavy breathing. Primarily, a benign tumor was not diagnosed correctly, and a patient was mistakenly diagnosed bronchial asthma. Complete examination in conditions of pulmonological department was performed 9 months after the complaints appeared. Instrumental methods including computed and magnetic resonance tomography of the neck and trachea, direct bronchoscopic visualization were very helpful for diagnosis. The final diagnosis was received after endoscopic resection of the neoplasm and histopathological study. Favorable clinical outcome was observed.

Conclusion

- Thus, the knowledge of clear diagnostic criteria of the upper and lower airway obstruction is necessary for administration of adequate therapy.
- Both acute stenosing laryngotracheitis and upper airway diseases of noninfectious genesis can cause asphyxia.
- When therapy administered according to up-to-date clinical recommendations is ineffective, the symptoms preserved and progressing require modern differential therapy to be applied.

Conflict of Interests

- E.G. Furman confirmed absence of conflicts of interests, on which it should be informed.
- E.A. Khuzina confirmed absence of conflicts of interests, on which it should be informed.
- N.N, Grymova confirmed absence of conflicts of interests, on which it should be informed.
- M.S. Ponomareva confirmed absence of conflicts of interests, on which it should be informed.

Bibliography

1. Blokhin BM. "Acute respiratory failure in children". *Medical Business* 4 (2008): 34-44.

2. Ferretti GR., *et al*. "Imaging of tumors of the trachea and central bronchi". *Thoracic Surgery Clinics* 20 (2010): 31-45.

3. Bharadwaj SC and Unruh HW. "Leiomyoma of the trachea". *The Annals of Thoracic Surgery* 93 (2012): 669-670.

4. Brown L M and Patterson AG. "Diagnosis and Management of Tracheal Neoplasms". *Cummings Otolaryngology Head and Neck Surgery and Oncology* VI (2015): 1751-1767.

5. Flint PW., *et al*. Lesperance M.M. (ed.) Cummings otolaryngology–head and neck surgery (2015): 168.

6. Dorenbusch AA. "Leiomyoma of the trachea: a case report". *AMA Archives of Otolaryngology* 61 (1955): 470-471.

7. Micheal C M., *et al*. "Advanced technologies for imaging and visualization of the tracheobronchial tree from computed tomography and MRI to virtual endoscopy". *Thoracic Surgery Clinics* 28.2 (2016): 127-137.

8. Zhang Z., *et al*. "Endobronchial leiomyoma in a patient presenting with chronic cough after treatment for pneumonia". *Chest* 146.4 (2014): 660.