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Case report
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TRACHEAL INFLAMMATORY MYOFIBROBLASTIC TUMOR IN A 3-YEAR-OLD BOY

INFLAMATORNI MIOFIBROBLASTNI TUMOR TRAHEJE KOD TROGODIŠNJEG DEČAKA

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Summary

Introduction. Inflammatory myofibroblastic tumors predominantly manifest in the lungs of children and young adults, with tracheal localization being very rare. Genetic alternations involving the anaplastic lymphoma kinase gene are identified in 50 to 70% of cases. A conclusive diagnosis relies on biopsy and histopathological analysis. Surgical resection stands as the primary treatment modality. **Case Report.** We present a case involving a 3-year-old boy with an inflammatory myofibroblastic tumor who had experienced recurrent wheezing attributed to respiratory infections since the age of eight months. Long-term therapy with budesonide and montelukast was initiated, which effectively managed his wheezing until the age of 3 years. Subsequently, despite ongoing medication, he began experiencing severe bronchial obstructions every month, necessitating repeated hospital admissions. At the age of 3 years and 8 months, he was admitted to our hospital due to persistent wheezing, prompting a bronchoscopy. During the procedure, a tumor-like mass was identified in the lower part of the trachea. Bronchoscopic removal of the tumor was performed, followed by cauterization of the remaining tumor tissue. Histopathological examination confirmed the presence of an inflammatory myofibroblastic tumor. **Conclusion.** Inflammatory myofibroblastic tumors are uncommon neoplasms associated with a borderline risk of malignancy. Given that the symptoms can resemble those of common childhood respiratory conditions, it is crucial to consider this diagnosis in cases of persisting wheezing despite standard therapy. In such instances, performing a bronchoscopy is necessary for accurate diagnosis.

Key words: Tracheal Neoplasms; Myofibroblasts; Bronchoscopy; Airway Obstruction; Respiratory Sounds; Child; Diagnosis, Differential

Sažetak

Uvod. Inflamatorni miofibroblastični tumori se najčešće javljaju u plućnom parenhimu dece i mladih odraslih, dok je njihova pojava u traheji veoma retka. Promene u genu za anaplastičnu limfomsku kinazu su potvrđene kod 50−70% pacijenata. Definitivna dijagnoza se postavlja na osnovu biopsije i patohistološke evaluacije. Osnovni vid terapije je hirurška resekcija tumora. **Prikaz slučaja.** Prikazan je inflamatorni miofibroblastični tumor traheje kod trogodišnjeg dečaka koji je imao epizode rekurentnog vizinga tokom respiratornih infekcija počev od osmog meseca života. Uvedena je terapija budesonidom i montelukastom i dečak je bio bez tegoba do treće godine života, nakon čega je imao je teške bronhoopstrukcije svakog meseca koje su zahtevale ponavljane hospitalizacije u lokalnoj bolnici. Kada je imao tri godine i osam meseci hospitalizovan je u našoj bolnici zbog perzistentnog vizinga. Sprovedena je bronhoskopija, opservirana je tumorska promena u donjem delu traheje. Načinjena je bronhoskopska ekstrakcija tumora uz kauterizaciju remnant tumora. Patohistološkim pregledom je potvrđen inflamatorni miofibroblastični tumor. **Zaključak.** Inflamatorni miofibroblastični tumori su retke neoplazme sa graničnim stepenom malignosti. Dijagnoza može biti maskirana simptomima i znacima najčešćih respiratornih bolesti u detinjstvu, poput rekurentnog vizinga kod predškolske dece. Bronhoskopiju treba uraditi kod svih pacijenata sa perzistentnim vizingom koji imaju neadekvatan klinički odgovor na standardnu terapiju.

Ključne reči: tumori traheje; miofibroblasti; brohnoskopija; opstrukcija disajnih puteva; respiratorni zvuci; dete; diferencijalna dijagnoza

Introduction

Inflammatory myofibroblastic tumors are rare neoplasms most commonly found in the lungs of children and adolescents [1, 2]. While they primarily occur in the lungs, they can also manifest in other extrapulmonary sites, though less frequently [2]. Additionally, inflammatory myofibroblastic tumor (IMT) is referred to by various terms in the literature, including plasma cell granuloma, inflammatory pseudotumor, inflammatory myofibrohistiocytic proliferation or inflammatory fibrosarcoma [3–5]. According to the World Health Organization's 2013 classification, myofibroblastic tumors are considered mesenchymal neoplasms of intermediate malignancy, exhibiting diverse histological and biological characteristics [1].

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Abbreviations

IMT – inflammatory myofibroblastic tumor
ALK – anaplastic lymphoma kinase
SMA – smooth muscle actin
COX-2 – cyclooxygenase-2

The etiology of IMTs remains unclear, but it is believed that these tumors arise from an aberrant inflammatory response triggered by various antigens. Changes in the anaplastic lymphoma kinase (ALK) gene have been observed in 50 to 70% of cases [6–8].

Clinical presentation of IMTs varies widely, with symptoms ranging from nonspecific laboratory and radiographic findings. A definitive diagnosis is established through biopsy and histopathological examination [6, 9, 10].

While surgical resection is the primary treatment modality, systemic therapies such as steroids, immunomodulators, radiotherapy, and chemotherapy may be considered for advanced or recurrent cases. Target therapy is an option for patients with confirmed ALK translocation [11–14].

Case report

The 3-year-old boy had a history of recurrent wheezing attributed to respiratory infections since the age of eight months and was managed with long-term asthma preventive therapy consisting of budesonide and montelukast. In addition to episodes of bronchial obstruction, the boy experienced periodic symptoms of allergic rhinitis. Further diagnostic evaluation revealed elevated IgE levels (305 IU/ml) and a positive skin prick test for tree pollen. Despite adhering to the regular asthma preventive therapy, the boy began experiencing severe bronchial obstructions on a monthly basis from the age of 3. These episodes necessiated repeated hospital admissions. As a consequence of persistent wheezing that did not respond to salbutamol inhalation, the boy was transferred from the local hospital to our institute.

Upon admission to our institute, the boy presented as afebrile, tachydyspnoic, and tachycardic, with a oxygen saturation of 96% on oxygen therapy delivered via a face mask at 6 liter per minute. Wheezing was heard on on chest auscultation, and a chest X-ray revealed perihilar inhomogeneous patchy infiltrates on the right side. Laboratory investigations showed leukocytosis (Le 26 G/L) and thrombocytosis (Tc 660 G/L), while blood gas exchange and electrolyte values remained within normal limits.

Initial management involved oxygen supplementation, inhaled salbutamol, corticosteroids (both inhaled and systemic), and antibiotic therapy. Persistent symptoms prompted further evaluation with bronchoscopy.

Fiberoptic tracheobronchoscopy under general anesthesia was initially performed by a respiratory pediatrician. During the procedure, a tumor-like mass was observed in the lower third of the trachea, situated on the lateral wall above the main carina and the right bronchus. The tumor occupied two-thirds of the trachea lumen and exhibited smooth surface that bled upon contact. It was attached to the trachea by a wide petiole. The fiberoptic bronchoscope was able to pass adjacent to the tumor, with the main carina appearing sharp, and the rest of the tracheobronchial tree showing no pathological changes. Subsequently, rigid tracheobronchoscopy was performed by an otorhinolaryngologist using a STORZ rigid bronchoscope (30 cm in length, size 5, with 0˚ telescope). The tumor change was visualized in the lower third of the trachea and removed using forceps. The dimensions of the tumor were 2 x 0.6 cm, and it exhibited a pale yellow color and firm consistency **(Figures 1 and 2).**

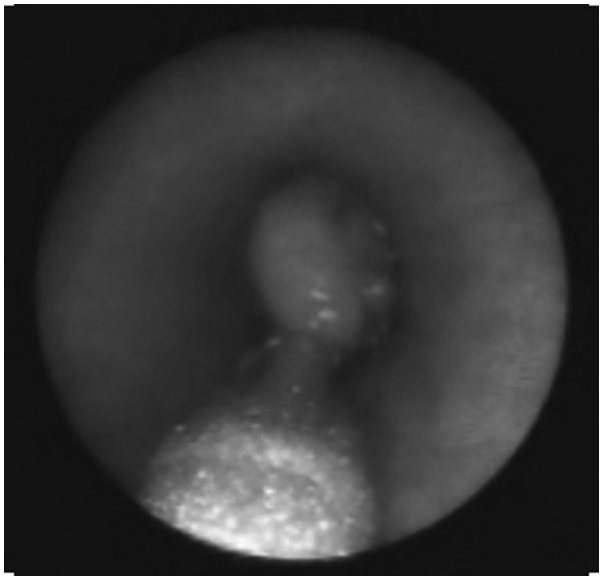

Figure 1. Bronchoscopic view showing the tracheal tumor arising from the lateral wall of the trachea
Slika 1. *Bronhoskopski prikaz tumorske promene lateralnog zida traheje*

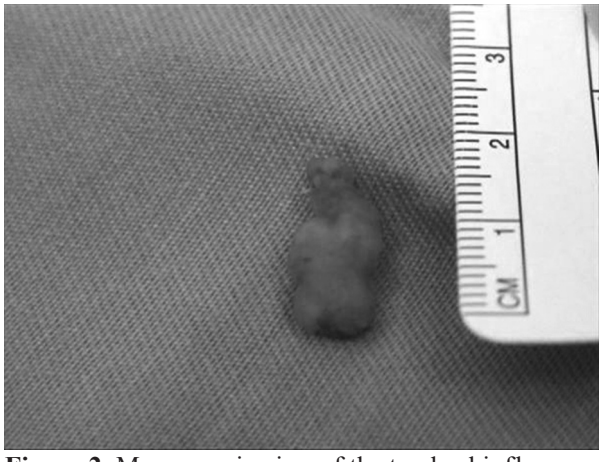

Figure 2. Macroscopic view of the tracheal inflammatory myofibroblastic tumor. The tumor surface is pale yellow color, appearing firm, measuring 2 x 0.6 cm.
Slika 2. *Makroskopski prikaz inflamatornog miofibroblastičnog tumora traheje. Površina tumora je svetložute boje, deluje čvrste konzistencije, dimenzija 2 x 0,6 cm.*

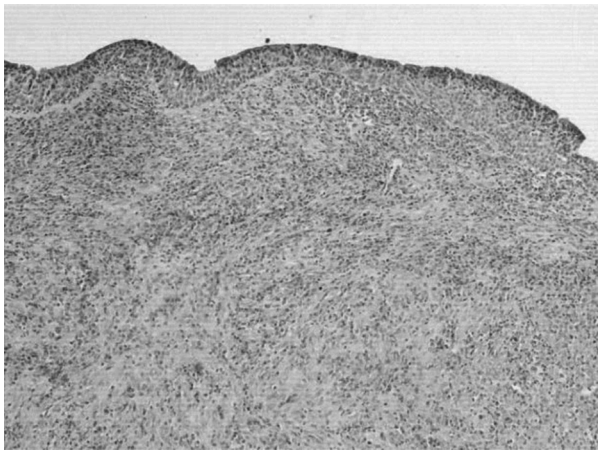

Figure 3. Photomicrograph of a mucosa with hyperplastic and metaplastic respiratory epithelium. In the lamina propria, there is diffuse infiltrate of plasma cells and lymphocytes among the bundles of spindle cells (HE x 50)
Slika 3. *Mikrografija mukoze sa hiperplastičnim i metaplastičnim respiratornim epitelom. U lamini propriji se nalazi difuzni infiltrat plazma ćelija i limfocita među snopovima vretenastih ćelija (HE x 50)*

The histopathological examination revealed that the surface of the tumor was partially covered by squamous epithelium and partially by respiratory epithelium. The epithelial surface showed signs of ulceration, with the bottom covered with granulocytes and fibrin. The lamina propria was replaced by a diffuse infiltrate of spindle cells arranged in short bundles in various directions. The cell nuclei appeared elongated and light, with visible cell nucleoli. Plasmacytoid and lymphocytoid cells were interspersed between the bundles of cells, while blood vessels were rare. Immunohistochemical staining revealed positivity to vimentin, smooth muscle actin (SMA), and ALK, while S100 and CKAE1/AE3 were negative. CD 68 and CD 45 showed focal positivity in single cells. These findings corresponded to an inflammatory myofibroblastic tumor **(Figures 3, 4 and 5).**

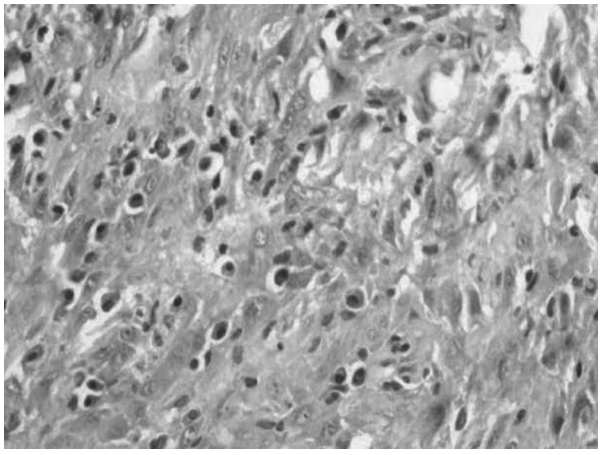

Figure 4. Detail of the tissue with plasma cells, lymphocytic cells, and numerous spindle myofibroblastic cells (HE x 400)
Slika 4. *Detalj tkiva sa plazma ćelijama, limfocitnim ćelijama i brojnim vretenastim miofibroblastičnim ćelijama (HE x 400)*

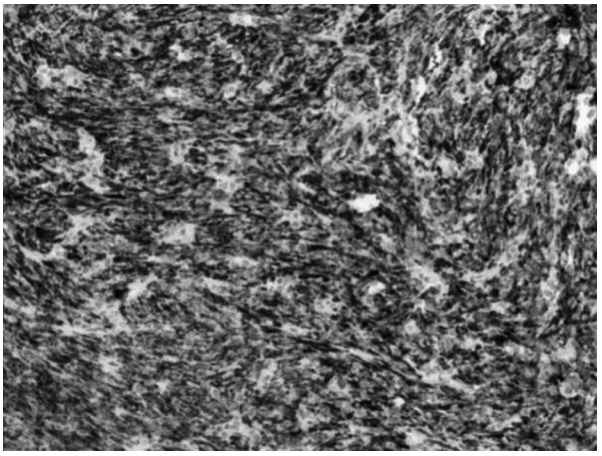

Figure 5. Immunohistochemical stain positive for expression of myofibroblastic cells of anaplastic lymphoma kinase ALK-1 protein (ALK x 200)
Slika 5. *Imunohistohemijsko bojenje pozitivno na ekspresiju miofibroblastičnih ćelija ALK-1 proteina anaplastične limfom kinaze (ALK x 200)*

Two weeks later, a follow-up tracheobronchoscopy revealed the presence of residual tumor attached to the site of the previously removed tumor in the trachea. It was observed to protrude above the surrounding smooth mucosa and was covered with blood. Subsequently, cauterization of the tumor remnant and its insertion site was performed. The next control tracheobronchoscopy was performed one month after the initial removal of the tumor and revealed the presence of scar tissue in the trachea at the site where the tumor had been removed previously. The final control tracheobronchoscopy was performed three months later, which showed no visible tumor recurrence.

Subsequently, the boy experienced periodic mild asthma exacerbations and symptoms of allergic rhinoconjunctivitis. The asthma preventive therapy was discontinued at the age of 8 years. Regular follow-up appointments with a pulmonologist were maintained, with the most recent pulmonary function checkup conducted at the age of 10. The boy remained asymptomatic, and his spirometry and body plethysmography results were within normal limits, as was his chest X-ray.

Discussion

Inflammatory myofibroblastic tumors (IMTs) were initially identified as primary lung tumors in 1939, and lung localization remains the most prevalent. However, IMTs can affect various organ systems, including the central nervous system, pelvic organs, retroperitoneum, gastrointestinal tract, orbit, and thyreoid gland [1–5]. According to the avail-

able data, IMTs account for 0.04% to 1.2% of all lung tumors, irrespective of age and sex [8]. IMTs are predominantly found in the lung parenchyma, with endobronchial tree localization accounting for approximately 5% of cases. However, its presence in the trachea is extremely rare, reported in only 2.7% of cases [3, 6–9]. In the case of our patient, the tumor was localized in the lower third of the trachea, on its lateral wall.

Traditionally viewed as reactive inflammatory lesions, IMTs are now classified as mesenchymal neoplasms of intermediate malignancy, as per the World Health Organization's 2013 classification. Their biological characteristics range from benign, locally aggressive, and recurrent forms to intermediate, potentially malignant lesions. Metastases are rare, occurring in less than 5% of cases [1, 5, 9]. Additionally, there have been reports of slow progression towards sarcoma [11]. Based on the findings from endoscopic evaluation of the respiratory tract in our patient, which did not show infiltration into surrounding structures, and the absence of recurrence in subsequent control bronchoscopies, it can be concluded that the inflammatory myofibroblastic tumor in this case exhibited a benign behavior.

The pathogenesis of IMTs remains uncertain. Several hypotheses suggest that IMTs may be associated with autoimmune or infectious mechanisms. It has been reported that in 30% of patients, IMTs have been linked to recurrent infections caused by Mycoplasma, Nocardia, Actinomycetes, and the Epstein–Barr virus. Other potential risk factors mentioned in the literature include corticosteroid use, surgery, trauma, or radiotherapy [3, 6–8]. In the case of our patient, the only identified risk factor was long-term use of inhaled steroids (budesonide) due to repeated bronchial obstruction prior to the diagnosis of IMT. Other risk factors were not present.

The activation of the immune system can cause genetic changes, and rearrangement in the ALK gene is the most common one. This gene is localized on the chromosome locus 2p23 and has been reported in 50 to 70% of cases. The activity of ALK kinase has been associated with apoptosis and has also been demonstrated in other tumors, such as small cell carcinoma of the lung and neuroblastoma [3, 7, 9]. Although some studies have shown better prognosis in tumors with ALK expression, the correlation between ALK expression and prognosis or risk of tumor recurrence is not clearly defined [3]. Other studies have found no difference in survival rates between ALK-positive and ALK-negative cases [1]. Some ALK-negative tumors are associated with ROS1 gen rearrangement. Recent studies have confirmed ETV6-NTRK3 fusion in ALK-negative pulmonary IMTs [3]. Activity of periostin, vimentin, desmin, SMA, CD 34, and S 100 are also noted [6–9]. In the case of our patient, immunohistochemical findings showed that the tumor was positive to vimentin, SMA, and ALK, and negative to S100 and CKAE1/AE3, while CD 68 and CD 45 showed focal positivity in single cells.

The diagnosis of IMT relies on the clinical presentation, laboratory and radiologic findings, endoscopic evaluation of the respiratory tract, and on histopathological verification [10–12]. The clinical presentation can vary widely, ranging from asymptomatic cases to those with severe respiratory symptoms, depending on the site of the primary tumor involvement. Some patients may also exhibit systemic manifestations such as fever, night sweats, or weight loss, often attributed to cytokine secretions [10–13]. Our patient exhibited persistent wheezing unresponsive to bronchodilator inhalation, and severe repeated bronchial obstructions requiring hospitalization. Laboratory abnormalities are uncommon, but when present, they may include findings such as anemia, thrombocytosis, elevated erythrocyte sedimentation rate, or hypergammaglobulinemia [3]. In the case of our patient, leukocytosis and thrombocytosis were observed, while other laboratory parameters were within normal limits. Radiological findings are non-specific, but imaging modalities such as chest X-ray or chest computed tomography can aid in determining the localization of the tumor [11, 12]. In the case of our patient, the chest X-ray revealed right perihilar inhomogeneous patchy infiltrates.

In children with persistent wheezing that is unresponsive to therapy, bronchoscopy evaluation is indicated to investigate potential causes of tracheobronchial obstruction. This evaluation aims to identify conditions such as foreign body aspiration, tracheal compression due to an anomalous blood vessel or other causes of tracheobronchial obstruction [12, 14]. In the case of our patient, who presented with persistent wheezing, bronchoscopy revealed intraluminal obstruction of the intrathoracic part of the trachea.

The final diagnosis of IMT is confirmed through histopathological findings [9–12]. The IMT typically consists of differentiated myofibroblasts and an inflammatory infiltrate, which includes plasma cells, lymphocytes, histiocytes, and eosinophilic granulocytes. Histologic patterns can vary, and different patterns may be observed in the same tumor [3, 6]. There are three histological subtypes of IMT: 1) the inflammatory subtype, which is characterized by a high concentration of mucus and inflammatory mediators; 2) the cellular subtype, primarily consisting of spindle myofibroblasts and inflammatory cells; and 3) the mixed type, with the predomination of collagen fibers [3,6]. In the case of our patient, the histopathological finding showed that the tumor's lamina propria was replaced by a diffuse infiltrate of spindle cells arranged in short bundles in various directions. Plasmacytoid and lymphocytoid cells were interspersed between the bundles of cells. These findings are consistent with the cellular subtype of IMT.

Macroscopically, IMTs typically present as lobular or multimodal lesions with a hard or rubbery consistency. These tumors predominantly appear as solitary lesions with dimensions ranging from 2 to 20 cm. Mul-

tiple nodular lesions are described in about one-third of cases [3]. In the case of our patient, the tumor was solitary, pale yellow in color, and possessed a firmer consistency. Its dimensions were measured at 2 x 0.6 cm.

In considering the differential diagnosis, several conditions should be taken into account. In cases of inflammatory leiomyosarcoma, cell nuclei are cigar shaped and arranged in regular fascicular patterns, while Reed Sternberg cells are observed in Hodgkin's lymphoma. Inflammatory fibroid polyps primarily manifest in the gastrointestinal tract. Imunoglobulin (Ig) G4 sclerosing disease typically affects older individuals. While some studies have reported the presence of IgG4-positive cells in IMTs, the extent of positivity of these cells and the IgG4/IgG ratio are typically lower compared to IgG4-related diseases [3].

The gold standard for the treatment of IMTs is complete resection of the tumor, either through interventional bronchoscopy or open lung surgery [13, 15, 16]. In our patient's case, bronchoscopic extraction of the tumor was successfully performed, along with cauterization of the tumor remnant and its insertion site during the control bronchoscopy.

Unresectable lesions require adjuvant therapies with chemotherapy, radiation therapy, COX-2 inhibitors, and steroids [13, 15, 17]. Corticosteroids have demonstrated efficacy in the treatment of some cases of unresectable lesions, although long-term use may lead to rapid proliferation of airway fibroblasts and tumor progression, possibly due to immunosuppression [18]. Celocoxib (a COX-2 inhibitor) and bronchoscopic argon plasma coagulation have emerged as treatment alternatives in recent years [17].

The discovery of ALK rearrangements has paved the way for targeted therapy with ALK inhibitors, with crizotinib being the most commonly used agent. Like most tyrosine kinase inhibitors, crizotinib acts on a wider scale than the target gene, inhibiting not only ALK but also ROS 1, and MET, which explains the positive treatment response in some ALK-negative tumors [13, 15, 16].

The prognosis is favorable following complete excision of the tumor. The five-year survival rate is approximately 90%, and the ten-year survival rate is nearly 70%. As local recurrences occur in 5 to 25% of patients, radiographic and bronchoscopic monitoring is recommended after the initial surgical treatment, as it was done in the case of our patient [8, 11–13].

Conclusion

Tracheal inflammatory myofibroblastic tumors are very rare neoplasms in children, with a borderline risk of malignancy. They can present with symptoms similar to the most prevalent respiratory illnesses, making diagnosis challenging. Therefore, bronchoscopy should be performed in persisting respiratory diseases, particularly wheezing, which do not improve with standard treatment.

The gold standard in the treatment of these tumors is radical surgery, while chemotherapy and radiotherapy may be considered for unresectable lesions, or recurrent tumors. Targeted therapy may be an option for the treatment of anaplastic lymphoma kinase positive inflammatory myofibroblastic tumors. Addressing the challenges associated with identifying new genetic rearrangements, refining therapies and application methods, as well as optimizing therapy duration represent key priorities to improved management and outcomes for affected patients.

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