A Rare Subglottic Capillary Hemangioma

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A 16-year-old boy was admitted to our emergency room with sudden respiratory distress. History revealed that he previously (from September to October 2009) suffered from progressive expiratory dyspnea, which he described as "something was floating up and down in his throat during respiration." Emergent laryngofiberscopy revealed a large, dark red, well-encapsulated, and pedunculated tumor located just below the glottis (Fig. 1). Moreover, the tumor was rising during expiration and sinking during inspiration. The tumor completely occluded the glottis during expiration, which could be fatal. Urgent tracheotomy was performed under topical anesthesia to keep the airway of the patient open and to prevent suffocation. Subsequently, the patient underwent neck computed tomography, which revealed a 1.5 × 1.0 cm oval subglottic tumor arising from the posterior wall of the upper trachea, approximately 2 cm from the vocal cords (Fig. 2). Three days after the tracheostomy, a complete resection was achieved through microlaryngoscopy under general anesthesia. Macroscopic evaluation of the surgical specimen revealed a 1.5 × 1.2 × 1 cm dark-red, homogeneous mass that originated from the soft tissue of the posterior tracheal wall. Microscopically, this mass was composed of a number of newborn capillaries, epithelioid cells, and fibrocytes. In addition, a considerable amount of black and brown particles were deposited in tissue mesenchyme (Fig. 3). Based on the observations mentioned above, the mass was diagnosed as capillary hemangioma. The patient was discharged 5 days after surgery without major complications. Furthermore, recurrence was not observed during the 5-year follow-up period.

Capillary hemangioma is a benign tumor composed of lobules of capillaries and surrounding fibrous tissue. It usually occurs in the skin and mucous membranes of children.[1] The oral and nasal cavities have been reported to be the common sites of involvement,[2] although it is rarely located in the trachea, especially the subglottic area.[3] No mechanism for the development of this tumor has so far been defined. Some etiological factors, including microtrauma, hormonal factors, and chronic inflammation stimulation, have been suspected to act in the pathogenesis.[4] In our case, however, the patient had no history of the factors mentioned above. Notably, the patient lived in a coal mine district for several years, which explained the unique pathological change of black carbon powder deposited in interstitial cells. We speculated that the continuing and chronic stimulation by coal dust might have been involved in the tumorigenesis. The absence of symptoms in the early stage and the large functional reserve of the tracheal lumen in adults often results in the delayed diagnosis. Diagnostic assessment with fiberoptic bronchoscopy and chest computed tomography are useful in planning therapeutic approach.[5] Although tracheal capillary hemangioma is benign, usually presenting no invasion; however, the constant growth

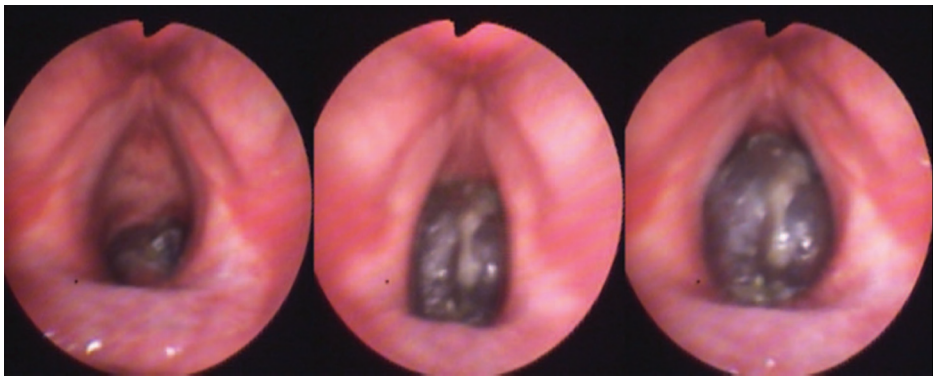

FIGURE 1. Laryngofiberscopy revealed a large, dark red, well-encapsulated, and pedunculated tumor located just below the glottis. The tumor rises during expiration and sinks during inspiration.

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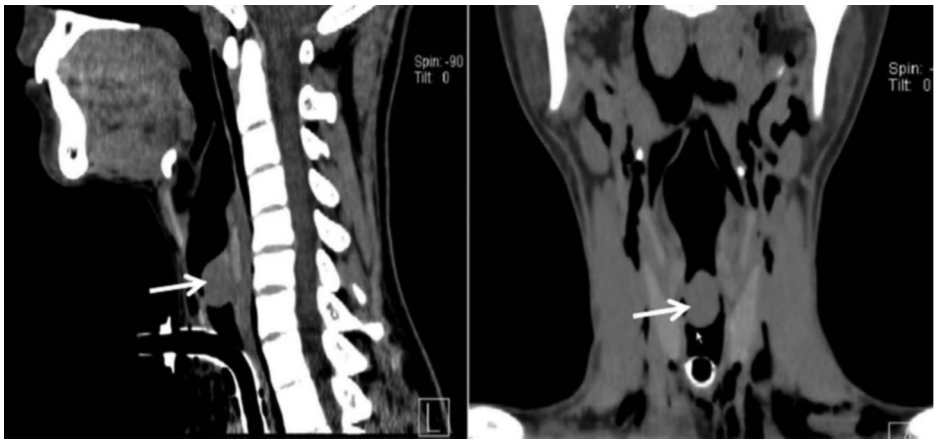

FIGURE 2. Neck computed tomography showed a 1.5 × 1.0 cm oval subglottic tumor arising from the posterior wall of the upper trachea.

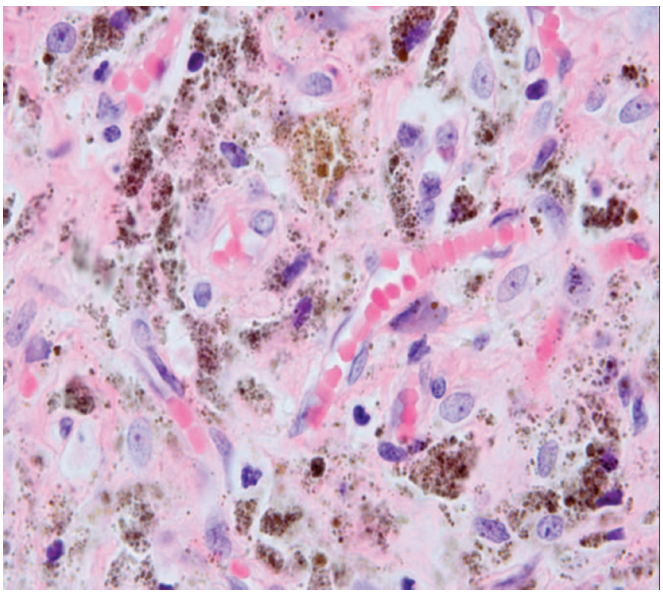

FIGURE 3. The mass was mainly composed of epithelioid cells, fibrocytes, and newborn capillaries. A considerable number of black and brown particles deposited in the tissue mesenchyme diffusely (hematoxylin and eosin stain stain, 200×).

may cause life-threatening airway obstruction. Several treatment protocols for tracheal tumor are available, such as laser ablation, electrocoagulation, and surgical excision.[6] Among them, complete surgical excision has been shown to provide definitive therapeutic effect with few recurrences.[7] In our case, the tumor was well encapsulated and connected to the posterior wall of the trachea by a fine pedicle. Thus, endoscopic complete resection via microlaryngoscopy was a preferential option with characteristics of less trauma and faster recovery.

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