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Laryngeal papillomatosis in a 3 year old infant: surgical and anesthesiological management

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Keypoints

This article describes a case of a 3-year-old infant with a history of hoarseness and persistent dysphonia caused by diffuse laryngeal papillomatosis. Urgent surgical intervention in microlaryngoscopy was performed with complete healing of the vocal folds and absence of papillomatous lesions after 1 month.

Abstract

Juvenile-Onset recurrent respiratory papillomatosis (JO-RRP) is the most frequent benign neoplasm in children with tendency to recur. Human Papilloma Virus (HPV) types 6 and 11 are involved in its etiology. Maternal transmission during the passage through the birth canal is the most frequent modality of infection. Clinical manifestations of JO-RRP may include dysphonia, dysphagia, hoarseness, cough and, in the most severe cases, respiratory distress. We report a case of a 3-year-old infant with a history of hoarseness and persistent dysphonia. Laryngoscopy showed diffuse papillomatous warts involving the glottic space and the false vocal cords.

An accurate preoperative anesthesiological evaluation was provided. Surgical intervention in microlaryngoscopy was performed under general anaesthesia.

Removal of the papillomatous warty growths in microlaryngoscopy was achieved, with significant improvement of the glottic space. No intraoperative or postoperative complications occurred, the patient resumed per os feeding on the same day and was discharged the second postoperative day. At the follow up visit at 1-month, important improvement of the voice quality was evident; laryngoscopy showed the absence of pathology; a strict follow up was planned in order to detect any recurrences.

Keywords

Laryngeal papillomatosis – papilloma virus – upper airways management - anesthesia in children – pediatric infection – vocal cords.

Introduction

Laryngeal papillomatosis is the most common benign laryngeal neoplasm in children with tendency to recur. When this condition occurs before the age of 12 years, it is called juvenile-onset recurrent laryngeal papillomatosis (JO-RRP) (*Ivancic 2018*). Its incidence has been estimated at 4.3 per 100,000 (*Derkay 1995*). It usually takes origin from HPV type 6 and 11 infection of the laryngeal epithelium (*Fasunla 2009).* Clinical manifestations of

laryngeal papillomatosis may include hoarseness, dysphonia, dysphagia and respiratory distress, requiring tracheostomy in the most severe cases. An early treatment is important to avoid respiratory complications (*Fasunla 2009*). It has been hypothesized that impaired T-cell mediated clearance of HPV infected cells may lead to the disease recurrence (*Vambutas 2000*). This article presents the surgical and anesthesiological management of a diffuse laryngeal papillomatosis in 3-year-old infant.

Case report

A 3-year-old patient was addressed for an otolaryngological evaluation at the ENT department because of persistent dysphonia. The symptom worsened in the previous months; no dysphagia or dyspnea were present. The patient had normal growth. At laryngoscopy, diffuse laryngeal warty growths involving true vocal folds, false vocal folds and anterior commissure with glottic space obstruction were evident (*figure 1*). Vocal folds motility was preserved, with incomplete glottic closure. Because of the severity of the obstruction, urgent surgical intervention in microlaryngoscopy was proposed.

Anesthesiology management started with the collection of an accurate familiar, physiological, pharmacological, remote and recent pathological anamnesis, which showed anything relevant; detailed objective examination was also performed, with particular attention to factors predictive of difficulty in airway management (the Colorado Pediatric Airway Score was used for a global assessment with a score of 6, which is predictive of a non-difficult intubation).

The patient was premedicated with midazolam per os 30 minutes before surgery and after ensuring an adequate monitoring (ECG, SpO2, NIBP, temperature, Bispectral Index with pediatric sensor, neuromuscular monitoring with TOF) and after an adequate preoxygenation with 100% oxygen in facial mask, general anaesthesia was inducted with administration of Fentanyl, Propofol and Rocuronium.

Orotracheal intubation was performed at first attempt using videolaryngoscope with paediatric blade (routine practice in our operating block since November 2021), which showed a full glottic view according to the Fremantle classification; a size 4 armored cuffed tracheal tube was used. Maintenance of general anaesthesia was achieved using Sevoflurane; Dexamethasone (0.1 mg/kg ev) was administered for the prevention of Postoperative Nausea and Vomiting (PONV) and of pharyngeal-laryngeal edema. After interrupting Sevoflurane delivery and after Sugammadex administration with TOFr > 90% extubation was performed. There were no anesthesiological complications.

At microlaryngoscopy, accurate removal of all the glottic papillomatous lesions was performed with attention to preserve healthy mucosa and the vocal ligament. Finally, important improvement of the

glottic space was achieved with a normal aspect of the vocal folds (*figure 2)*.

There were no postoperative complications; the patient resumed per os nutrition the same day and was discharged on the second post-operative day. At 1-month follow up evaluation, fiberoptic laryngoscopy showed complete healing of the vocal folds and absence of papillomatous lesions. The voice improvement was evident. A strict follow up was suggested in the next months in order to promptly detect any recurrences.

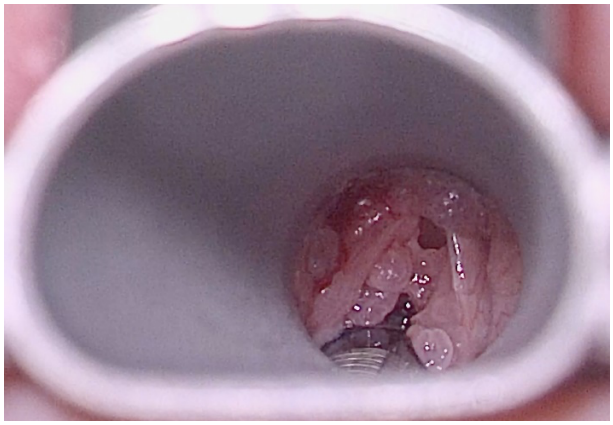

Figure 1. *The intraoperative view at microlaryngoscopy shows diffuse warty growths involving the true vocal cords and false vocal cords with significant reduction of the respiratory space.*

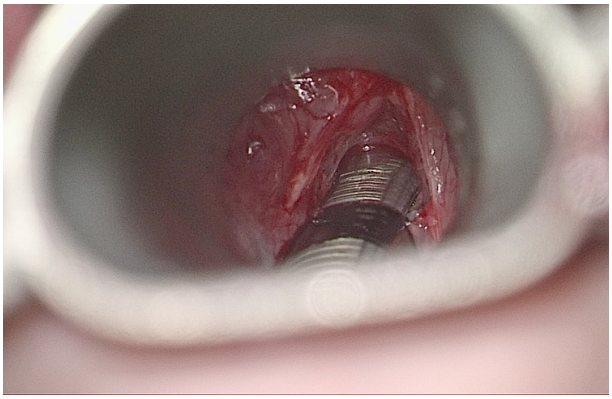

Figure 2. *At the final aspect, the papillomatous lesions have been removed and improvement of the glottic space is evident. The vocal ligament has been preserved.*

Discussion

Laryngeal papillomatosis is the most common benign laryngeal neoplasm in children; its presentation may mimic other lesions in the larynx (*Coope 2006*). Maternal transmission of the infection occurs during the passage through the birth canal; however, blood transmission has been described too (*Vergechik 2010*). The clinical presentation in children is usually not specific for the disease hence high index of suspicion is required to make diagnosis. Its clinical course may include frequent disease recurrence, significant morbidity, and occasional mortality (*Zacharisen 2006, Tasca 2006*). Furthermore, a 2% malignant degeneration incidence has been observed in recurrent respiratory papillomatosis patients (*Nebesio 2001*). According to the literature, the duration of illness before presentation to the hospital varies between 5 weeks and 3 months (*Fasunla 2009*). In the most advanced cases, tracheostomy may be necessary; tracheostomized children present increased morbidity; moreover, tracheostomy could promote the spread of the disease beyond larynx (*Mgbor 2005*). The treatment of laryngeal papillomatosis include removal of laryngeal papillomatous growth in microlaryngoscopy; some authors report the use of alfa-interferon, acyclovir, cidofovir injections, CO_2 laser, microdebrider and argon plasma coagulation (*Dancey 2000*, *Bergler 1997*). Because anesthesia and surgery share the same airway, establishing an effective airway for both anesthesia and surgery is very difficult. Anesthesiological management of laryngeal papilloma needs special attention as the lesion can obstruct already narrow pediatric airway, in addition anxiety and respiratory distress in the child makes it more challenging to the anesthesiologist. The goals of anesthesia are to provide adequate ventilation, provide the vocal cord relaxation, to avoid trauma, laryngospasm and to provide good surgical access. The special measures necessary for securing the airway include thorough preoperative airway evaluation, the appropriate choice of the anesthesia induction method and careful intraoperative and postoperative airway management. In fact, pediatric patients with laryngeal papillomatosis may present poor intolerance to anoxia, frequent relapse, scar contracture and throat antrum stricture after multiple operations (*Bo 2011*). The introduction of the quadrivalent HPV vaccine, Gardasil, which targets low-risk HPV types 6 and 11 and high risk HPV types 16 and 18 may improve the management of JO-RRP (*Carifi 2015*). A recent systematic review of seven studies investigated the role of quadrivalent HPV vaccination for secondary prevention of RRP. All seven case reports or cohort studies treating active RRP with quadrivalent HPV vaccination reported an increased interval between surgeries or decreased recurrence (*Dion 2017*).

JO-RRP is a rare disease which may cause severe upper respiratory airways obstruction and tends to recur frequently. Multiple treatment modalities have been proposed in the past years in order to reduce the risk of recurrence, however a definitive cure is still not available. Strict cooperation between the anesthesiologists and otolaryngologist is cardinal during the operative management of such patients. Increased awareness of this condition in pediatricians and general practitioners may allow early recognition and improve treatment outcomes.

Conclusion

In this article we report a case of anesthesiological and surgical management of a diffuse laryngeal papillomatosis in an infant. An early treatment was essential to avoid

respiratory complications and an accurate removal of all the glottic papillomatous lesions under general anesthesia was achieved. A strict follow up was planned in order to detect any recurrences.

Ethical Committee approval

The approval of the regional ethical committee is not requested for case reports.

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Conflict of interest

The authors declare no conflict of interest.

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