Awake fiberoptic nasal intubation in an infant with a malignant rhabdoid tumor occupying the oral cavity: a case report

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Abstract
Malignant rhabdoid tumors develop in infants up to 1 year old. These tumors are a very progressive pediatric cancer that is difficult to treat. We experienced a case involving anesthetic management for tracheostomy of an infant with a high risk of airway occlusion due to a rapidly growing malignant rhabdoid tumor on his tongue. The patient was successfully intubated using an endotracheal tube with a 3.0-mm inner diameter; this was accomplished by awake nasal intubation using a flexible rhinolaryngoscope with a 2.2-mm outer diameter. Awake nasal fiberoptic intubation is an alternative choice for difficult airway management in infants.

Keywords: Anesthesia induction, awake nasal fiberoptic intubation, malignant rhabdoid tumor, oral cavity tumor, pediatric difficult airway management

Background
Malignant rhabdoid tumor (MRT), which was first reported in 1978, is one of the most progressive pediatric cancers and develops in infants up to 1 year old [1]. We experienced a case involving anesthetic management for tracheostomy in an infant with a high risk of airway occlusion due to a rapidly growing MRT on his tongue. Awake fiberoptic nasal intubation was carried out using a flexible rhinolaryngoscope with an ultra-thin diameter.

Case presentation
A 5-month-old, 4.9-kg male infant developed a rapidly growing tumor on his tongue and was hospitalized 3 months after birth. Chemotherapy was started after admission; 2 weeks later, pathological analysis revealed an MRT. Although the tumor decreased in size with early chemotherapy, it began to grow aggressively again, and his oral cavity was almost completely occupied by the tumor 8 weeks after admission (Figure 1A). He exhibited difficulty swallowing saliva, and preoperative magnetic resonance images showed that he was at a high risk of airway occlusion (Figures 1D and 1E). Emergent tracheotomy by otolaryngology under general anesthesia was planned. However, the tumor occupying his oral cavity almost completely blocked the approach route for tracheal intubation via the mouth. Because we were not sure whether any airway devices can improve the unexpected airway occlusion and there was a high risk of “cannot ventilate, cannot intubate” (CVCI) during asleep anesthesia induction, we decided to keep spontaneous breathing of the infant and perform awake nasal tracheal intubation, instead of taking alternative anesthetic considerations, such as sedative induction using intravenous ketamine or dexmedetomidine, or asleep intubation utilizing volatile anesthetics. First, without any sedation, by using a suction catheter, topical anesthesia (approximately 1 mL of 4% lidocaine/epinephrine, 50ng/mL) was administered to the nasal mucosa, the lower pharynx, and the larynx to control the airway reflexes. Next, we advanced an uncuffed 3.0-mm-ID endotracheal tube into the lower pharynx through his nose, and inserted a flexible rhinolaryngoscope (ENF-XP®; Olympus) with a 2.2-mm outside diameter (OD) and 30-cm working length (Figures 2A and 2B upper), checked the location of the larynx, and advanced the tip of the scope into the trachea. Then, we successfully advanced the endotracheal tube into his trachea using the scope as an insertional guide-line for the nasal intubation (Figures 2C and 2D). Anesthetic induction was then carried out with sevoflurane inhalation and rocuronium intravenous injection, and tracheotomy and
Figure 1. Malignant rhabdoid tumor in the oral cavity and preoperative magnetic resonance (MR) images.
A: The tumor in the oral cavity 1 day before the tracheostomy.
B: The tumor in the oral cavity 2 days after the tracheostomy.
C: Three months after the tracheostomy.
D: Preoperative MR image (sagittal view).
E: Preoperative MR image (horizontal view).

Broviac catheter insertion were performed. The surgery was completed in about 1 hour (Figures 2E and 2F). One day after the surgery, the tumor mass had increased in size and was extruded to the outside of the oral cavity, suggesting that the patient would have developed complete airway occlusion if the surgery had been delayed (Figure 1B). The tumor gradually regressed with subsequent chemotherapy (Figure 1C). Three months later, resection of the residual tumor on his tongue was performed under general anesthesia. The patient's condition was still well controlled at the time of this writing.

Discussion
MRT is a very aggressive tumor that occurs mainly in children [1]. In 1989, Weeks et al., [2] reported 111 cases of MRT. They showed that although it was originally described as a variant of Wilms' tumor, which is primarily a kidney tumor, many types of rhabdoid tumors outside the kidney have been reported in various tissues including the liver, soft tissue, and central nervous system. In addition, they reported that regardless of location, all MRTs are highly aggressive, have a poor prognosis, and tend to occur in children less than 2 years of age [2]. In our infant patient, a tumor originated from his tongue and aggressively increased in size within his oral cavity during several weeks, almost occluding his airway. Only two infant cases of oral MRT have been previously reported. First, in 1997, Pizer et al., [3] reported a case involving a large MRT arising from the mouth of an infant. In their case, the oral tumor was associated with a mass in the posterior cranial fossa. Second, Patron et al., [4] reported an MRT of the tongue in a 10-day-old boy who developed severe respiratory distress, had an aggressive clinical course, and died 17 days after the initial diagnosis.

In 1999, White et al., [5] described the clinical, pathologic, and immunohistochemical features of a widely disseminated tumor with a rhabdoid phenotype in nine infants under 3 months of age. Five neonates had tumor evident at birth, two of which had placental metastases, and the average survival duration following diagnosis was <6 weeks. In four cases, the dominant mass involved the head and neck region, and in two cases, the primary mass was paraspinal. Genetic information was available from eight of the nine cases. Karyotype analysis revealed abnormalities of chromosome band 22q11-12 in three of six tumors, and in situ hybridization studies or molecular studies demonstrated 22q11.2 deletions in all five cases [5]. More recently, an abnormality in the hSNF5 (INI1) gene in 22q11.2 was demonstrated [6]. Because MRT grows very aggressively, if the tumor is associated with the patient's airway as in our case, a decision regarding the surgical procedure is required before critical obstruction and respiratory distress occur. Note that although previous reports have implied an aggressive disease course and poor prognosis of this tumor, the original oral tumor in our case was diminished by postoperative chemotherapy at least at the time point 1 year after the first admission.

With the recent development of flexible fiberoptic scopes...
with small diameters for pediatric patients, flexible fiberoptic scope-assisted nasal intubation has become another tool for managing difficult airways, even in infants [7-9]. In such cases, an ultra-thin intubation fiberscope with a 2.2-mm OD and 60-cm working length is useful for intubation of an endotracheal tube with a 2.5-mm ID and 3.0-mm ID as reported previously [10,11]. During oral fiberoptic laryngoscopy and tracheal intubation, supralaryngeal airway devices such as laryngeal mask airways can provide an excellent direct channel to the vocal cords while allowing for either spontaneous or controlled ventilation and oxygenation, simultaneously shielding the bronchoscope from secretions and bleeding. However, an adequately large mouth opening and working space in the oral cavity are required to place a supralaryngeal airway device before insertion of an endotracheal tube. Therefore, awake nasal intubation is sometimes valuable when the patient’s mouth opening is limited and when an obstruction exists in the patient’s oral cavity. In general, it is suggested that the OD of the fiberoptic bronchoscope should be 1 mm smaller than the ID of the endotracheal tube (Figure 3, Table 1) [7] while fiberoptic bronchoscopes as small as 2.2-mm in OD, such as the Olympus laryngoscope LF-P®, Olympus Rhinolaryngoscope ENF-XP®, and Pentax nasopharyngo-laryngoscope FNL-7RP3®, can be used for endotracheal tubes as small as 2.5-mm in ID (Figure 2B lower). Disadvantages of fiberscopes with a small diameter previously included a limited field of vision and no instrumental channel, but all of the above-mentioned fiberscopes have relatively superior optical performance and break through 120-130° up/down angulation capabilities (Table 1). In the present case, we used the Olympus ENF-XP®, which has a 2.2-mm OD and 30-cm working length (Figure 2A). The major difference among the Olympus LF-P®, Olympus ENF-XP®, and Pentax FNL-7RP3® is their working length; the Olympus LF-P® has a 60-cm working length, while the Olympus ENF-XP® and Pentax FNL-7RP3® have a 30-cm working length. Our impression is that for nasal intubation in infants, fiberscopes with a 30-cm working length are more advantageous than those with a 60-cm working length in terms of the ability to handle the flexible shaft of the scope.

Conclusions
In summary, in our experience with this case involving an infant with a difficult airway due to a large malignant rhabdoid tumor in the oral cavity, an appropriate choice was awake nasal fiberoptic intubation of an endotracheal tube with a 3.0-mm ID using an ultra-thin fiberscope with a 30-cm working length.

Table 1. List of commercially available fiberoptic scopes and their outer diameter and working length.

| OD (mm) | Vendor | Name | Product type | Working length (cm) | Channel (Port) | Tip deflection up/down |
|---------|--------|------|--------------|---------------------|---------------|------------------------|
| 2.2     | Olympus| Laryngoscope | LF-P | 60 | - | 120°/120° |
| 2.3     | Pentax | Intubation scope | FI-7P/7BS | 60 | - | 130°/130° |
| 2.4     | Pentax | Bronchoscope | FB-8V | 60 | + | 180°/130° |
| 3.0     | Pentax | Intubation scope | FI-9BS | 60 | + | 130°/130° |
| 3.1     | Olympus| Laryngoscope | LF-DP | 60 | + | 120°/120° |
| 3.4-3.5 | Pentax | Intubation scope | FI-10P2/10BS | 60 | + | 130°/130° |
| 3.5     | Pentax | Bronchoscope | FB-10V | 60 | + | 180°/130° |
| 3.6     | Pentax | Nasopharyngo Laryngoscope | FNL-10RP3/10RBS | 30 | - | 130°/130° |
| 4.1-4.2 | Olympus| Intubation/Laryngoscope | LF-2/GP | 60 | + | 120°/120° |
| 4.3     | Pentax | Intubation scope | FI-13P/13BS | 60 | + | 130°/130° |
| 4.9     | Pentax | Bronchoscope | FB-15V | 60 | + | 180°/130° |
| 5.1-5.3 | Pentax | Nasopharyngo Laryngoscope | FNL-15RP3 | 30 | - | 130°/130° |

OD: Outer diameter
length (Olympus Rhinolaryngoscope ENF-XP®).

Additional files

Supplement video 1
Supplement video 2

List of abbreviations
ID: Inner diameter
MRT: Malignant rhabdoid tumor
OD: Outer diameter

Competing interests
The authors declare that they have no competing interests.

Authors' contributions

| Authors' contributions                        | AT | HK | FT | MS | YK | TS | YN |
|----------------------------------------------|----|----|----|----|----|----|----|
| Research concept and design                  | -- | -- | -- | -- | -- | ✓  | ✓  |
| Collection and/or assembly of data           | ✓  | ✓  | ✓  | ✓  | -- | -- | -- |
| Data analysis and interpretation             | ✓  | -- | -- | -- | ✓  | -- | -- |
| Writing the article                          | -- | -- | -- | -- | -- | ✓  | -- |
| Critical revision of the article             | -- | -- | -- | ✓  | -- | -- | ✓  |
| Final approval of article                    | ✓  | ✓  | ✓  | ✓  | ✓  | ✓  | ✓  |
| Statistical analysis                         | -- | -- | -- | -- | -- | ✓  | ✓  |

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