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Cryotherapy and balloon dilatation for subglottic stenosis in a neonate: a case report

Johanes Edy Siswanto[1,2]*, Akira Prayudijanto[1], Muhammad Iqbal[1], David D. Putra[3], Istifa Amalia[3], Rifan Fauzi[1], Christophorus AS Pamungkas[4]

[1]Department of Pediatrics, Harapan Kita National Women and Children Health Centre, West Jakarta, Indonesia, [2]Faculty of Medicine, Pelita Harapan University, Tangerang, Indonesia, [3]Department of Pulmonology and Respiratory Medicine, RSUP Persahabatan, Jakarta, Indonesia, [4]Faculty of Medicine, University of Indonesia, Jakarta, Indonesia

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ABSTRACT

Cryotherapy and balloon dilatation are combined treatments for subglottic stenosis (SGS) in newborns. Cryotherapy involves freezing the affected area with extremely low temperatures, which can result in tissue destruction. Balloon dilatation is a technique in which a balloon is inflated in the narrow subglottic area to widen the airway. We reported a case of SGS in a neonate who was treated with a combination of cryotherapy and balloon dilatation to contribute to the growing body of evidence of the therapy and provide information for clinicians in the care of the vulnerable patients. A 22-day-old female baby, who weighed 2,600 gr at birth, was transferred to our hospital from another facility. We conducted cryotherapy on the granulation tissue area until it was released, then inflated the balloon for 5 cycles (30 sec each) with a pressure of 5-6 atm. After cryotherapy and balloon dilatation, the patient was examined using direct laryngoscopy, which revealed subglottic stenosis up to 9.5 cm from the outer border of the mouth. To maintain airway patency for the next 24 hr, the patient was intubated with an endotracheal tube number 3.5. The patient's condition improved after this intervention, and they no longer required oxygen therapy support. The patient was discharged without the need for supported oxygen. In conclusion, to manage the challenges associated with post-intubation subglottic stenosis (PI-SGS), a comprehensive approach that includes a reliable referral system, thorough bronchoscopy training, and a multi-disciplinary team is essential.

ABSTRAK

Krioterapi dan dilatasi balon merupakan kombinasi tindakan pengobatan yang dilakukan pada stenosis subglotik bayi baru lahir. Krioterapi adalah prosedur medis untuk membekukan dengan suhu dingin yang ekstrem pada area yang terkena, sehingga dapat menyebabkan penghancuran jaringan. Sedangkan dilatasi balon adalah teknik menggelembungkan balon di area subglotik yang sempit untuk memperluas jalan napas. Dilaporkan sebuah kasus subglotik stenosis pada bayi baru lahir yang diterapi dengan kombinasi krioterapi dan dilatasi untuk dapat berkontribusi pada semakin banyak bukti tentang terapi subglotik stenosis pada bayi baru lahir dan memberikan informasi berharga untuk klinisi yang terlibat dalam perawatan pasien yang rentan ini. Seorang bayi perempuan, berusia 22 hari dengan berat saat lahir 2.600 gr, merupakan kasus rujukan ketergantungan oksigen yang berasal dari rumah sakit lain. Kami melakukan tindakan krioterapi pada area jaringan granulasi sampai terlepas, kemudian diikuti dengan dilatasi balon selama lima siklus (30 detik) dengan tekanan 5-6 atm. Selanjutnya dilakukan eksplorasi dengan laringoskopi langsung sampai batas luar mulut, 9,5 cm, dan didapatkan stenosis subglotik pasca tindakan krioterapi dan dilatasi balon. Untuk mempertahankan saluran pernapasan selama 24 jam berikutnya, kami mengintubasi pasien dengan pipa endotrakeal nomor 3.5. Kondisi pasien membaik, terlepas dari ketergantungan oksigen dan segera dapat dipulangkan. Simpulan, untuk menangani *PI-SGS*, perlu dilakukan pendekatan komprehensif yang mencakup sistem rujukan yang baik, pelatihan bronkoskopi yang menyeluruh, dan memiliki tim multidisiplin.

Keywords:
bronchoscopy;
balloon dilatation;
cryotherapy;
neonate subglottic;
stenosis

*corresponding author: edyjo15@yahoo.com

INTRODUCTION

Congenital subglottic stenosis (SGS) is a condition that typically affects infants under three months of age. It is characterized by a narrowing of the space below the vocal cords in the throat, leading to symptoms such as difficulty breathing, biphasic or inspiratory stridor, and recurrent croup. Acquired SGS, on the other hand, can result from procedures like frequent intubations, extended endotracheal intubation, or the use of a large tube, and it is a serious long-term complication caused by posttraumatic fibrosis of the larynx.[1,2]

Treatment options for SGS include observation, injection therapies, surgical procedures, dilation, stent placement, laryngotracheal reconstruction (LTR), or tracheostomy. Observation with routine at-home or in-clinic spirometry may be considered for patients with mild symptoms.[3]

Cryotherapy and balloon dilatation are two treatment modalities that have been explored for subglottic stenosis in neonates. Cryotherapy involves applying extreme cold to the affected area, leading to tissue destruction. On the other hand, balloon dilatation is a technique in which a balloon is inflated in the narrowed subglottic area to widen the passage. Balloon dilation is often considered the initial treatment for acquired subglottic stenosis, boasting over a 90% success rate in improving symptoms and reducing the need for additional surgical procedures.[4] However, some providers prefer to start treatment with injections before proceeding to dilation procedures. The success of balloon dilation is strongly linked to a shorter duration, lower initial grade of stenosis, younger patient age, and the absence of tracheotomy.[5] We reported a case of SGS in a neonate who was treated with a combination of cryotherapy and balloon dilatation. Through this case report, we aim to contribute to the expanding body of knowledge about the treatment of neonatal subglottic stenosis and provide important insights for physicians caring for these patients.

CASE

A female baby, who was 22-day-old and weighed 2,600 gr at birth, was referred to our hospital from another facility. She was on mechanical ventilation and had been in the neonatal intensive care unit (NICU) for 2 wk. While on non-invasive ventilation, she started experiencing inspiratory stridor while breathing. During the first bronchoscopy examination, we noticed that the pharynx was collapsing and the epiglottis was moving backwards during inhalation. The arytenoids appeared slightly swollen, and the vocal cords were symmetrical both in static and dynamic positions. There was subglottic stenosis, which was too narrow for an OD 3.1 mm scope to pass through, with 95% obstruction (Myers-Cotton grade III). We performed resection of stenosis using a Holmium laser (8 kJ) until it was partially opened, followed by cryotherapy and balloon dilatation for 5 cycles of 15 sec each. However, upon re-evaluation, we discovered that there was multilevel stenosis. We then attempted intubation using an ETT number 3.5 with a scoping guide, but it was unsuccessful. Finally, we planned intubation with direct laryngoscopy and ETT number 2.5 cm, with the outer border of the mouth measuring 9 cm. During expiration, the trachea's posterior wall gave way, and the carina opened, completing the procedure. The result was subglottic stenosis and pharyngolaryngotracheomalacia (FIGURE 1C).

During the following week, we repeated the same procedure. We observed the following: pharyngeal collapse on inspiration, posterior

movement of the epiglottis during inspiration, symmetrical vocal cords in static and dynamic states, and visible granulation tissue in the subglottic area. The granulation tissue was obstructing the airway by 50-60% (Myers-Cotton grade II), as shown in FIGURE 2A. We performed cryotherapy on the granulation tissue area until it was released, followed by inflating the balloon for five cycles (30 sec) with a pressure of 5-6 atm, as shown in FIGURE 2B. After reevaluation, the stenosis area was almost 90% open (proximal trachea position deviation), and we attempted intubation with ETT number 3.5 with a scope guide (intranasal). Still, the ETT could not pass through the nasal area. We successfully intubated the patient with direct laryngoscopy to the outer border of the mouth, 9.5 cm, and the result was stenosis subglottic post-cryotherapy and balloon dilatation, as shown in FIGURE 2B. To maintain the patency of the airway for the next 24 hr, we intubated the patient with an endotracheal tube number 3.5. The patient's condition improved, and they were weaned off oxygen therapy and discharged without the need for supported oxygen.

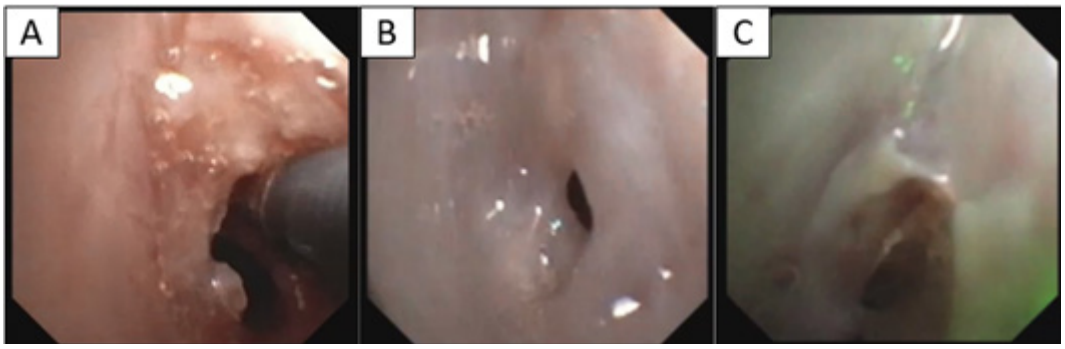

FIGURE 1. A) The initial procedure used cryotherapy and ballooning. (B) Before the first therapy, it revealed subglottic stenosis with 95% obstruction (Myers-Cotton grade III). (C) The first cryotherapy and ballooning treatment resulted in subglottic stenosis and pharyngolaryngo-tracheomalacia.

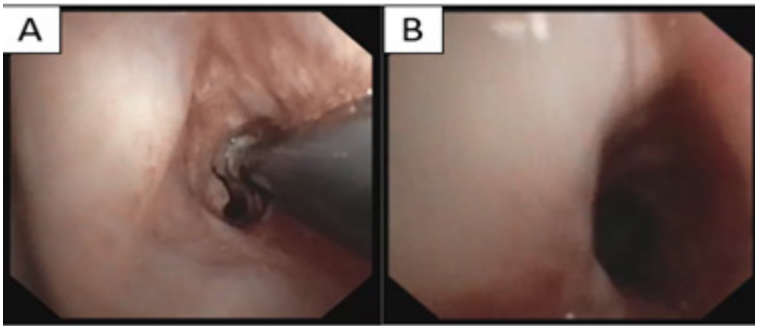

FIGURE 2. A) The second procedure used cryotherapy and ballooning. B) The second cryotherapy and ballooning treatment resulted in the area of stenosis is almost 90% open.

DISCUSSION

The subglottis is an anatomical region that extends inferiorly to the bottom border of the cricoid cartilage, below the level of the vocal folds. It is surrounded by the thyroid cartilage and the cricothyroid membrane superiorly, and the structures inferior to the trachea. The cricoid is the only complete cartilaginous ring in the airway, and due to its fixed diameter, even subtle edema or scarring can lead to a potentially life-threatening airway condition.[6]

Congenital SGS is a condition that occurs in infants under three months of age and is characterized by a narrowing of the space below the vocal cords in the throat. Symptoms of SGS include difficulty breathing, biphasic or inspiratory stridor, and recurrent croup. Conversely, frequent intubations, extended endotracheal intubation, or the use of a large tube can all lead to acquired SGS. This type of SGS is a serious long-term complication that results from posttraumatic fibrosis of the larynx.[1,2]

Although SGS is a serious condition, its incidence has been decreasing since the late 1990s. According to a 2001 study by Walner et al., the incidence of SGS at the moment is most likely between 0 and 2%. Less than 4% was observed in research published after 1983, while less than 0.63% was recorded in studies published after 1990. To diagnose SGS, laryngoscopy and bronchoscopy are necessary to evaluate the airway and rule out other conditions, such as subglottic cysts or hemangiomas. Acquired SGS is typically more severe than the congenital type.[7]

Managing subglottic stenosis (SGS) in children is a complex and challenging aspect of pediatric care, requiring a collaborative approach involving pediatric otolaryngologists, anesthesiologists, pulmonologists, and intensive care specialists. It's crucial to promptly address respiratory distress in infants, especially when there's increased breathing effort despite normal oxygen saturation levels, as this could indicate an impending crisis. In such cases, quick decisions regarding the safest way to secure the airway are vital. The operating room provides the safest environment for securing the airway. If endotracheal intubation proves difficult, direct laryngoscopy (DL) or rigid bronchoscopy can be performed there. Preparation for an emergency tracheostomy should be made as a backup if securing the airway is unsuccessful. It's important to note that cricothyroidotomy is not recommended for children due to the size and collapsibility of the airway.[8]

Direct laryngoscopy and bronchoscopy are utilised in the operating room to identify and evaluate the severity of SGS. Prior to any procedure, all patients undergo direct laryngoscopy and bronchoscopy to assess the condition of the larynx and the extent of tissue stenosis. The severity of tissue stenosis is categorized using the Cotton-Myer grading method. Grade I signifies a stenosis of up to 50%; grade II, 51% to 70%; grade III, 71% to 99%; and Grade IV, no lumen present.[9]

In this case, we are utilizing endoscopic techniques, including balloon dilatation, laser therapy, and cryotherapy, to treat our patients. These methods are less invasive than surgical procedures such as tracheostomy, which is of particular importance for our young patients. Endoscopic therapies offer numerous benefits, such as reversibility, shorter hospital stays, and less surgical time.[9] There is no one-size-fits-all endoscopic therapy option; instead, each patient's needs are unique. Stent implantation, T-tube installation, and argon plasma coagulation are options for children with post-intubation SGS.[10]

Thirty-three pediatric patients, averaging the age of 31 mo, received endoscopic balloon dilatation.[11] Among these patients, 15 had acute SGS, 18 had

chronic SGS, 4 had Myers-Cotton grade I, 9 had Myers-Cotton grade II, and 20 had Myers-Cotton grade III. Thirteen patients with acute SGS and eighteen individuals with chronic SGS had positive results. The success rate for grade I was 100%, grade II had an 88.9% success rate with five patients experiencing re-stenosis, and grade III had a 60% success rate with 14 patients experiencing re-stenosis.[11]

Complications such as the formation of granulation tissue and re-stenosis are often observed after bronchoscopic interventional treatments like electrosurgery, balloon dilatation, T-tube or stent implantation, and laser therapy. Injection of local medicine has been considered an effective therapy to stop granulation tissue formation and re-stenosis, while its efficacy is still questionable.[12]

The holmium laser is a safe and effective option for precise ablation and hemostasis in paediatric circumstances because of its cutting ability, which is similar to that of the CO_2 laser, and its coagulation capacity, which is similar to that of the Nd: YAG laser. It is the greatest laser alternative for paediatric interventional treatment since it can remove scar tissue and granulation effectively.[11] With less chance of resulting in cartilaginous damage than laser therapy, cryotherapy is an additional safe and efficient bronchoscopic technique for treating airway blockage.[11] Jiao *et al.*[13] presented the multifold advantages of the holmium laser with cryotherapy. Their cohort suffered no major complications. Those patients with preoperative tracheostomies underwent successful decannulation, and no patients in the cohort required new tracheostomy placement.[13] After receiving the treatment, all 16 patients showed immediate improvement in their signs and symptoms, 15 of them finished the entire course of treatment and achieved a clinical cure. Among the advantages were a noticeable improvement in endoscopic lumen diameter, respiratory symptoms, hypoxemia, voice quality, and swallowing function. A Cotton-Myer Grade IV patient had clinical improvement; nonetheless, they were lost to follow-up.[13]

Post-intubation Subglottic Stenosis in Children: Case Discussion

Several recent studies support the effectiveness and safety of combining cryotherapy with holmium laser or other bronchoscopic techniques in the treatment of pediatric subglottic stenosis (SGS). Jiao *et al.*,[14] reported that holmium laser therapy followed by cryotherapy resulted in complete clinical cure in 15 out of 16 pediatric patients, including those with severe SGS, with no need for new tracheostomies and no major complications. Zhang *et al.*,[15] also demonstrated the success of holmium laser cryoablation in combination with inhaled budesonide in infants, showing significant airway improvement and reduced restenosis risk. Imran Yılmaz *et al.*,[16] confirmed cryotherapy's role in managing dual SGS in complex infectious settings, while still avoiding open surgery. Lastly, Hosna *et al.*,[17] emphasized the benefit of repeated cryotherapy sessions and early intervention, with a favorable safety profile and good airway remodeling outcomes in neonates. These findings underscore the growing role of minimally invasive modalities in neonatal airway management and align with the approach used in our case.

CONCLUSION

Early diagnosis is crucial for timely treatment of PI-SGS. Physicians must also understand the risks of intubation in neonates to overcome the challenges associated with PI-SGS. A comprehensive approach includes a reliable referral system, thorough bronchoscopy training, and a multidisciplinary team.

RECOMMENDATION

Early diagnosis of PI-SGS is essential recommended so the patient may receive treatment as early as possible. Physicians have to know the risks of intubation in neonates, as PI-SGS may happen, as they are very prone to developing stenosis. Therefore, a good referral system, bronchoscopy training, and multi-disciplinary approaches are important to handle the problems due to PI-SGS.

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ABBREVIATIONS

Atm : Atmosphere
Cm : Centimeter
ETT : Endotracheal Tube
kJ : Kilo Joule
NICU : Neonatal Intensive Care Unit
OD : Ocular Dextra
PI-SGS : Post-Intubation Subglottic Stenosis
SGS : Subglottic Stenosis

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