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Case Report

Irradiated homologous costal cartilage grafts for single-stage open airway reconstruction in severe subglottic stenosis for children under the age of one

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ABSTRACT

We present two cases of premature infants with Cotton-Myer grade 3 subglottic stenosis where endoscopic balloon dilation failed. In an attempt to avoid tracheostomy, both patients underwent open single stage anterior graft larnygotracheoplasty (LTP) with irradiated homologous rib and temporalis fascia grafts.

After postoperative reconstruction, the airway was resized to grade 1 in one child, and to grade 2 in the other. One child remained free of stridor post-operatively and avoided the need of a tracheostomy tube. The other child ultimately required a tracheostomy, though she is now in the process of being decannulated.

Homologous costal cartilage grafts in open anterior airway LTP may be a reasonable alternative to tracheostomy in young patients with advanced SGS.

1. Introduction

Acquired subglottic stenosis (SGS) is a common occurrence in preterm infants. The subglottis, as the narrowest part of the neonatal airway, is prone to injury from intubation leading to scarring and eventual stenosis. A spectrum of SGS exists, from mild croup-like symptoms to significant stridor and respiratory arrest, necessitating the need for definitive airway management [1–4].

Historically, the standard treatment for clinically severe SGS has been tracheostomy, though this is associated with significant morbidity and, consequently, alternative treatments have been explored. Surgical expansion of the laryngotracheal complex through a single stage laryngotracheoplasty has shown to be a viable alternative, and may allow for the avoidance of a tracheostomy in select cases [5]. Costal cartilage remains the mainstay reconstructive material, though many autologous cartilage grafts have been described including auricular, nasal septal, hyoid and thyroid ala cartilage [6–9]. Disadvantages of autograft costal cartilage include donor site morbidity, the risk of pneumothorax, limited tissue availability and increased operative time.

As an alternative to autologous costal cartilage, irradiated homologous costal cartilage, has emerged, having been successfully used in a wide variety of cosmetic and reconstructive procedures [10]. Unlike an autologous source, it is readily available, requires decreased operative time and avoids donor site morbidity. To the best of our knowledge, only one group has published their experience with the use of irradiated homograft costal cartilage for LTP as part of a double stage reconstruction. We build on this previous literature with two cases of single stage open airway reconstruction in severe subglottic stenosis for children under one year of age.

2. Methods

Two ex-premature female infants with stridor and respiratory distress and grade III anterior subglottic stenosis were evaluated at Al Jalila Children's Specialty Hospital. In both cases, their subglottic stenosis failed to improve after endoscopic balloon dilation and the families were reluctant to proceed with tracheostomy. Pre-existing comorbidities excluded either patient from being a candidate for autologous rib graft harvest. Both children were ex-premature (27 and 24 weeks premature), had failure to thrive and significant cardiopulmonary risk factors. As such, they may not have had the reserve to tolerate possible complications from rib graft harvest. Following an informed consent process, a decision was made to proceed with single stage laryngoplasty using a homologous cartilage graft.

A commercially available homograft was obtained from the Tutoplast Tissue Sterilization Process (RTI Surgical Holdings -

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Marquette, USA). The preparation methodology is well described elsewhere [11]. In brief, it is a graft cleaning and preservation process using solvent dehydration. Lipids are first removed from all grafts in an ultrasonic acetone bath. A series of alternating hyperosmotic saline and distilled water baths are then employed as well as a hydrogen peroxide solution. Grafts then undergo a final acetone wash and are placed in sterile packaging. Donor history screening and laboratory testing are compliant with the U.S. Food and Drug Administration (FDA) regulations and American Association of Tissue Banks (AATB) Standards [11]. The Tutoplast process has been commercially available for over 30 years [11]. Contaminant cells are lysed and washed away during the process, exposing the RNA/DNA and enveloped and non-enveloped viruses. The process has been proven to be effective in eradicating more than 12 log of infectivity [11].

A single stage anterior graft LTP was completed by the senior author using the standard surgical and anesthetic technique. In order to mimic the perichondrium, a similar irradiated homograft of temporalis fascia from the same company was sutured to the inner surface of the irradiated cartilage homograft with vicryl sutures positioned laterally. The graft was then sutured to the trachea with either a 5–0 or 6–0 prolene using interrupted sutures. Roughly 4–5cm of adult rib cartilage was available for each case. Handling of the homograft is comparable to an autograft, though it is somewhat softer and as a result slightly more difficult to carve.

2.1. Case 1

This was a 10-month-old ex-27-week preterm infant with a history of severe infant respiratory distress syndrome, bronchopulmonary dysplasia, gastro-esophageal reflux disease and developmental delay. She had a history of prolonged intubation in the neonatal period with multiple failed extubating attempts. With respect to her airway, she had a grade 3 anterior subglottic stenosis without any glottic involvement that failed endoscopic dilation. Her intraoperative course was unremarkable and she was stented with a 4.0 uncuffed endotracheal tube (ETT). Four months following reconstruction, her anterior graft was well integrated (Fig. 1). At last follow-up (15 months post LTP), the child remains well with no audible stridor and a grade 1 subglottis.

2.2. Case 2

This was a 5-month-old born at 24 weeks with numerous comorbidities including: perinatal asphyxia, grade 3 intra-ventricular hemorrhage with hydrocephalus, bronchopulmonary dysplasia, gastro-esophageal reflux disease, retinopathy of prematurity, previous patent ductus arteriosus ligation, anemia and failure to thrive. Pre-treatment she had a grade 3 subglottic stenosis without any glottic involvement. Her preop ETT size was 2.5 uncuffed and she was stented with a 3.5 uncuffed ETT.

Her intraoperative course was unremarkable. However, her post treatment course was complicated by the appearance of an airway hemangioma, distal to the site of reconstruction and in the posterolateral trachea. This hemangioma was not previously detected by preoperative direct laryngoscopy and tracheoscopy, likely because it was stented for a prolonged period by the existing 2.5 ET tube. It was ablated using the CO2 laser without recurrence.

Following reconstruction, she had no stridor and her subglottis was the same diameter as trachea. Unfortunately, with every subsequent upper respiratory tract illness, she would require intubation for pulmonary toilet and ventilation, but always extubated and discharged home without oxygen and without stridor when recovered. Ultimately a decision was made to proceed with tracheostomy, to avoid repeated admissions and intubations. The surgery was uncomplicated and postoperatively, she did well, gaining 5kg in 6 months. She is currently in the process of getting decannulated.

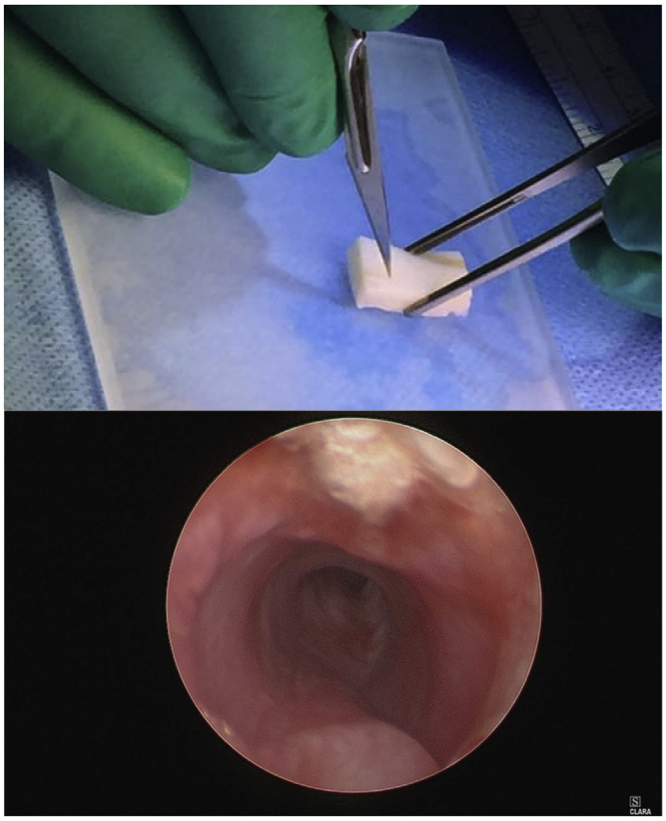

Fig. 1. Case 1. (A) Preparation of the rib cartilage graft. (B) Anterior cartilage graft in situ at 4 months post-op.

3. Discussion

Single stage LTP has been shown to be successful in the majority of infants under 6 months with clinically significant subglottic stenosis [5]. Surgical expansion of the subglottic space with an autologous costal cartilage graft remains the current standard at many pediatric institutions. Cartilage has been found to be superior to other materials, like bone, due to lower rates of resorption, ease of use (i.e. 'carvability') and viability without a vascular pedicle [12]. That said, donor site morbidity, limited tissue availability and increased operative time have motivated groups to search for alternatives.

Animal models have been used to evaluate the use of irradiated cadaver costal cartilage in LTP. The cartilage had significant resorption and did not gain wide uptake [13]. However, cadaveric cartilage has experienced widespread use in cosmetic rhinoplasty and other reconstructive head and neck procedures [10,14–16]. In a review of over 350 patients undergoing irradiated homologous cartilage grafts for rhinoplasty, the complication rate was equivalent to autologous sources [17]. On the other hand, a recent study did note higher resorption rates in irradiated homologous costal cartilage over autologous costal cartilage in a cohort of 63 patients undergoing augmentation rhinoplasty [18]. Additionally, it has been postulated that irradiated cadaveric cartilage may be more suspectable to infection [19]. These are considerations that should be discussed as part of an informed consent process.

An alternative to homograft would be anterior thyroid ala cartilage [8,20]. Like homologous sources, it has the advantage of having only one surgical site and a graft, though in our experience, the thyroid ala is often too small to adequately enlarge the airway. Tissue engineered cartilage grafts also demonstrate good potential, though have only been explored in animal models at present [21].

To date, only one previous study reported on results of irradiated cadaveric cartilage in LTP. Smith and Pereira described their use of

homologous costal cartilage in five tracheostomy dependent children as part of a delayed double stage reconstruction [22]. Only minor degrees of resorption were noted, and they were successfully able to decannulate 4 of 5 patients. Our experience is similar in our two patients with single stage LTP, though the extrusion of prolene in one of our patients would have us favor a dissolvable stitch, like polydioxanone, in the future. In hindsight, case 2 may have been better suited for a delayed/two-stage reconstruction. That said, at the time of the decision, the parents were adamant that a tracheostomy should not be performed, unless it was in the context of a lifesaving intervention.

4. Conclusion

Homograft costal cartilage is a successful alternative to autograft in LTP. It reduces operative time and negates the donor site morbidity of many autologous sources. The graft appears effective, but the long-term resiliency in the setting of LTP remains unknown.

Declaration of competing interest

No financial disclosures/conflict of interest to declare.

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