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CASE REPORT

Slide tracheoplasty without cardiopulmonary bypass for the treatment of long and complex congenital stenosis: case report and review of the literature

Traqueoplastia slide sin bypass cardiopulmonar para el tratamiento de la estenosis congénita larga y compleja: reporte de caso y revisión de la literatura

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Abstract

Congenital tracheal stenosis is an entity that causes acute airway obstruction and is associated with high morbidity and mortality due to diagnostic delay and its accompanying comorbidity. Slide tracheoplasty is the procedure of choice that traditionally requires the use of cardiopulmonary bypass to facilitate the tracheal reconstruction technique. There are no national reports of this tracheoplasty. With a modification to the original description, the surgical technique is described and a case report executed without the use of cardiopulmonary bypass. A review of the literature is proposed to guide the comprehensive approach and treatment.

Keywords: *Tracheoplasty. Slide tracheoplasty. Congenital tracheal stenosis. Bronchoscopy.*

Resumen

La estenosis traqueal congénita es una afección que causa obstrucción aguda de la vía aérea y asocia unas altas morbilidad y mortalidad ante el retraso diagnóstico y por sus comorbilidad acompañante. La traqueoplastia slide es el procedimiento de elección y requiere tradicionalmente el uso de bypass cardiopulmonar para facilitar la técnica de reconstrucción traqueal. No existen reportes nacionales de esta traqueoplastia. Con una modificación a la descripción original se describe la técnica quirúrgica y se reporta un caso sin uso de bypass cardiopulmonar. Se propone una revisión de la literatura que permita orientar un abordaje y un tratamiento integrales.

Palabras clave: *Traqueoplastia. Traqueoplastia slide. Estenosis traqueal congénita. Broncoscopia.*

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Introduction

Congenital tracheal stenosis (CTS) is an urgent condition that causes acute airway obstruction and is associated with high morbidity and mortality due to delayed diagnosis and accompanying comorbidities. Long and complex CTS is characterized by the presence of complete tracheal rings along the stenotic segment, which determines a fixed narrowing of the tracheal lumen that manifests with stridor and respiratory distress in early infancy. Cardiovascular abnormalities occur in 69% of cases, with vascular anomalies such as pulmonary artery sling (ring) being the determinants of prognosis and survival. Bronchoscopy is the diagnostic standard and the main strategy for surgical planning and follow-up[1]. Since its first description by Tsang et al.[2] in 1989, slide tracheoplasty is considered the procedure of choice for long CTS and traditionally requires the use of cardiopulmonary bypass (CPB) to facilitate the tracheal reconstruction technique. There are no national reports of this slide tracheoplasty for the treatment of CTS. With a modification to the original technique, the surgical procedure is described, and a case of slide tracheoplasty performed without the use of CPB is reported. A review of the literature is proposed to guide the treatment of this congenital airway emergency.

Case report

A 3-month-old male with trisomy 21 and intrauterine growth retardation, with biphasic stridor and respiratory distress at birth. Intubation was achieved with a 3.5 mm endotracheal tube (ETT) that stopped in the neck without advancing distal to the carina. Intubated for 1 month and hospitalized for 2 months for pneumonia, he was discharged. He presented to the emergency department with severe respiratory distress and was stabilized again with a 3.5 mm endotracheal intubation with the tip of the ETT at neck level. Echocardiography showed a 3 mm atrial septal defect. He presented with pulmonary hypertension of 50 mmHg and a forced expiratory volume of 65%, without structural heart disease and a state of heart failure due to an increase in airway resistance. Evaluation was performed with an initial flexible bronchoscope of 2.8 mm, finding fixed tracheal stenosis in the second tracheal ring, extensive and infundibular with complete rings, which did not allow passage of the equipment. Rigid tracheal dilation was decided, and the airway was secured with a 3 mm ETT up to the carina. With improvement of hemodynamic conditions, a computed tomography scan with airway reconstruction was performed, revealing long, complex tracheal stenosis with inspiratory collapse in the left bronchus. With the patient stable, he underwent slide tracheoplasty at 4 months of age (Fig. 1).

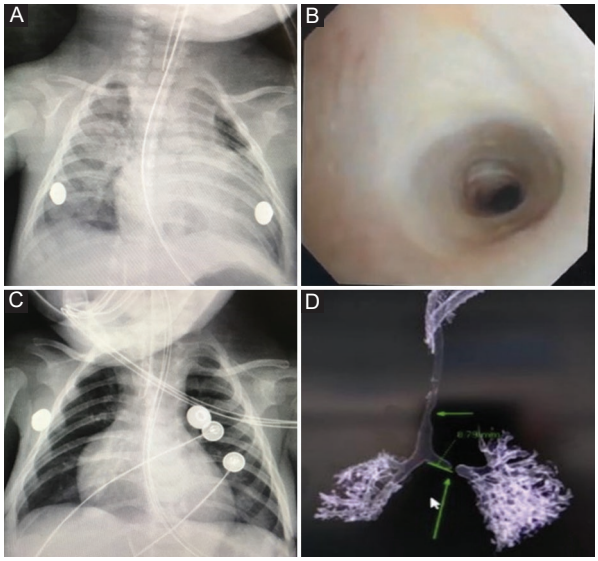

Figure 1. A: *infant with stridor and respiratory distress with a 3 mm endotracheal tube (ETT) that stops at cervical airway level.* **B:** *presence of critical infundibular stenosis with complete rings starting from the second tracheal ring, confirming the diagnosis of congenital tracheal stenosis (CTS) by bronchoscopy.* **C:** *distal advancement of a 3 mm ETT to the carina after rigid tracheal dilation, improving hemodynamic and respiratory conditions.* **D:** *long-segment CTS with inspiratory collapse in the left bronchus on a tomographic reconstruction of the airway.*

Surgical technique

With intubation using a 3 mm ETT at the carina, a median sternotomy with cervical extension was performed, followed by dissection with preservation of supra-aortic vessels to expose the entire trachea up to the carina and bronchial origins. The trachea was transected at the midpoint of the longitudinal stenosis, and 2 complete and central tracheal rings were resected, including the most critical narrowing, for pathological study. A medial and anterior longitudinal incision was performed in the distal or inferior tracheal slide, allowing ventilation with a 3 mm ETT with a cuff (this change of the original technique allowed reconstruction without conventional CPB throughout the entire surgery). The proximal or superior end was prepared as a posterior slide with a medial and

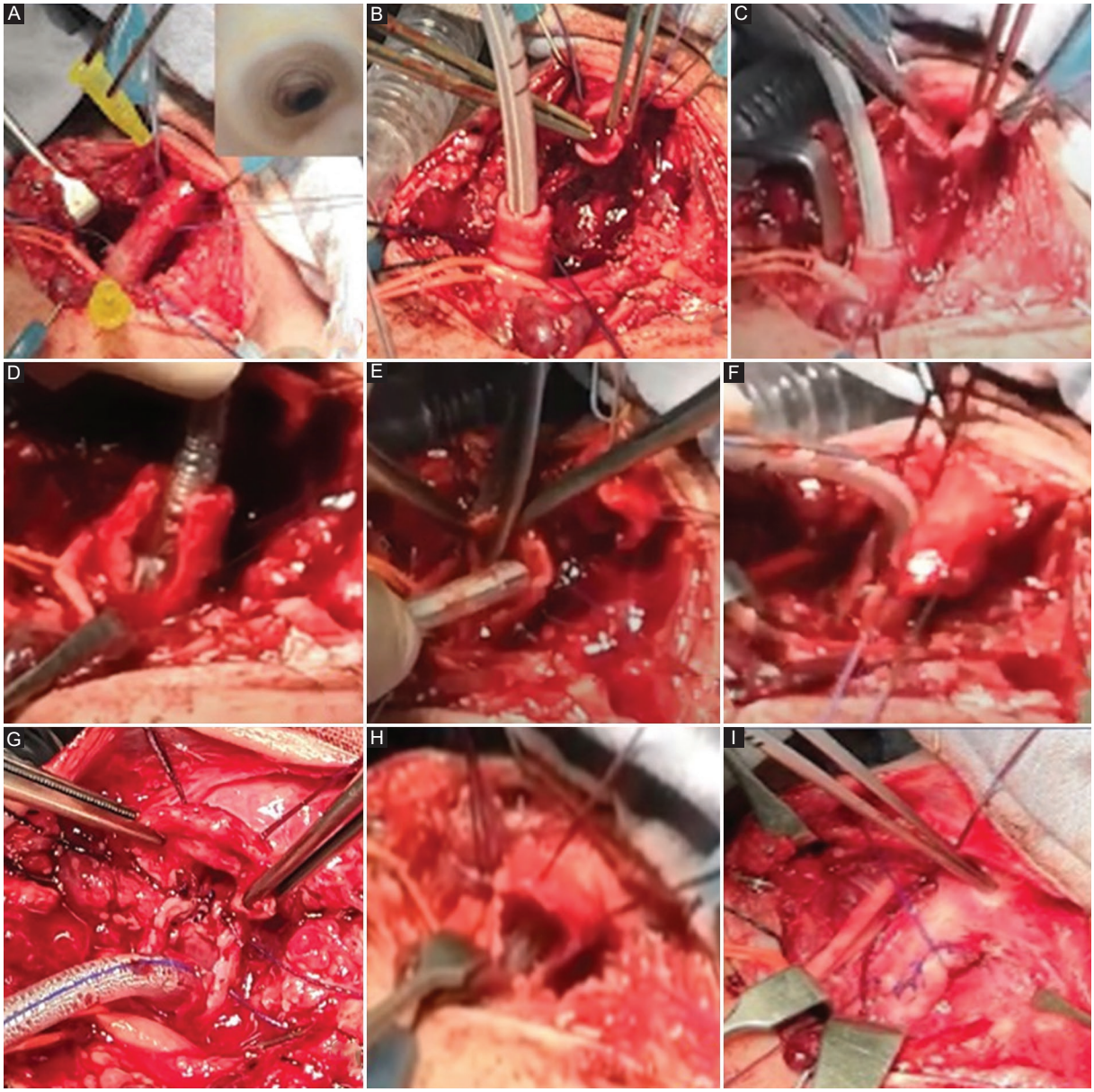

Figure 2. A: *median and cervical sternotomy with preservation of supra-aortic vessels exposing the entire trachea. The extent of congenital tracheal stenosis (CTS) is determined by intraoperative bronchoscopy, and the ends of the critical stenosis are marked with 2 needles. Note the severe reduction in the caliber of the trachea.* **B:** *transection of the CTS at the longitudinal midpoint between the 2 previously marked cephalic and caudal needles. The diagnostic presence of complete tracheal rings with the absence of membranous trachea in both tracheal ends is confirmed, and ventilation is initiated with a 3 mm endotracheal tube (ETT) in the distal tracheal end.* **C:** *the proximal tracheal end is prepared as a posterior slide: medial longitudinal incision in the posterior trachea extending to the second stenotic tracheal ring.* **D:** *preparation of the distal tracheal end as an anterior slide: medial longitudinal incision of the anterior tracheal wall extending to the left bronchial origin over the 3 mm cuffed ETT inflated for intraoperative ventilation.* **E:** *the triangular ends of both slides are spatulated to achieve a tongue-like arrangement.* **F:** *tension-free sliding of both overlapping slides is confirmed.* **G:** *posterior tracheal anastomosis starting at the apex of both slides. Ventilation through the distal slide continued throughout the surgery.* **H:** *anastomosis and reconstruction of the anterior neotrachea. A 3.5 mm ETT appropriate for the patient's age is advanced.* **I:** *final arrangement of the slide tracheoplasty (the dissecting forceps points to the cricoid cartilage). Integrity of tracheal reconstruction confirmed under bronchoscopic vision.*

posterior longitudinal incision. The edges of both tracheal slides were spatulated to achieve a tongue-like arrangement. The approximation of both overlapping ends was confirmed without tension and adequate irrigation. Anastomosis was completed by overlapping both preformed tracheal slides or ends with absorbable 4-0 Vicryl suture. After completing the posterior anastomosis, a 3.5 mm uncuffed ETT was advanced;

the anterior anastomosis, completed; and the integrity of the slide tracheoplasty, confirmed with a brief extubation and intraoperative bronchoscopy showing an acceptable increased caliber of its lumen and advancement of the equipment towards the pulmonary segmentation. Secretions were aspirated, and the patient reintubated with a 3.5 mm ETT above the neocarina. He was transferred to the ICU without drains (Fig. 2). In the postoperative follow-up, successful extubation was performed on day 7, being discharged on day 40. He experienced readmissions for septic events associated with his immunodeficiency, requiring 3 uneventful endotracheal intubations with an age-appropriate ETT. At 1 year of age, he presented with a granuloma and 50% concentric and fibrous tracheal stenosis at the apex of the tracheoplasty, refractory to endoscopic management. Tracheoplasty was performed uneventfully for resection of 2 tracheal rings in the middle third of the trachea with end-to-end anastomosis. Evident growth in length and post-slide airway lumen was noted, with an intact airway on bronchoscopy and asymptomatic at the 2-year follow-up (Fig. 3).

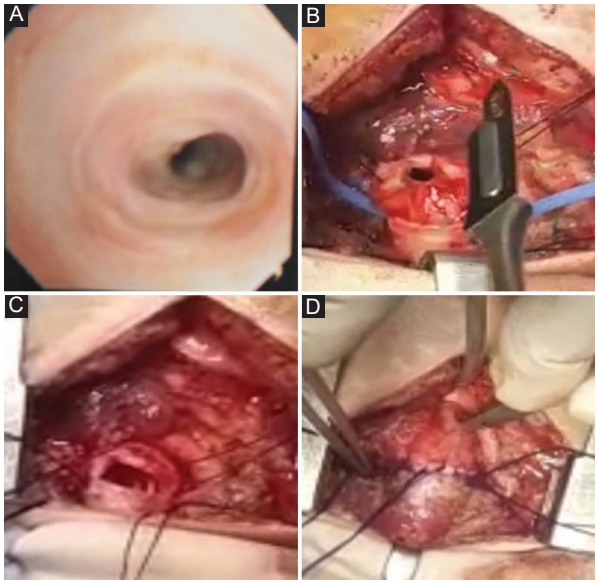

Figure 3. A: *acquired intrinsic stenosis in the post-slide neotrachea refractory to endoscopic management. Note the increase in tracheal lumen.* **B:** *resected stenotic sixth tracheal ring.* **C:** *tracheoplasty with tension-free end-to-end anastomosis with an age-appropriate endotracheal tube.* **D:** *post-slide neotrachea with a diametrical and longitudinal increase greater than the congenital tracheal stenosis observed in figure 2A.*

Discussion

Numerous techniques have been described for the surgical reconstruction of CTS: tracheal resection and end-to-end anastomosis, augmentation tracheoplasty with autologous pericardial or costal cartilage patch, and slide tracheoplasty. Since its first description by Tsang et al.[2] in 1989, slide tracheoplasty has been accepted as the preferred reconstruction technique, with greater prognostic and surgical advantages over other methods: definitive and rigid tracheal reconstruction, with airway stability with vascularized and preserved tracheal tissue that impacts a tracheobronchial reconstruction without excessive tension, doubles the tracheal circumference and increases the tracheal area up to 4 times, early extubation with a reduction in the formation of granulation tissue and with potential tracheal growth in the postoperative period and follow-up[3]. Bronchoscopy is the most important diagnostic strategy, as it confirms, as in our patient, the characteristic presence of complete cartilaginous tracheal rings, and the absence of membranous trachea; furthermore, it allows deciding the surgical strategy in the presence of associated aerodigestive malformations, such as pulsatile compression of the airway by an accompanying vascular ring and acquired lesions resulting from trauma during endotracheal intubation and the scenario of previous endoscopic or surgical procedures. Bronchoscopy assisted therapeutic rigid dilation and secured the airway to improve the patient's hemodynamic and preoperative conditions. When distal endoscopic evaluation of the airway is not possible due to the critical narrowing of the tracheal lumen, computed tomography with 3D reconstruction of the airway allows grading the extent and severity of the compromised airway; similarly, it rules out the presence of associated cardiovascular malformations functionally evaluated during echocardiography. Both elements complement the choice of the most appropriate surgical strategy[4].

Half of patients with CTS have an accompanying pulmonary artery sling-type vascular ring, so its preoperative recognition is mandatory to conventionally perform vascular reimplantation of the left pulmonary artery to the trunk portion of the main pulmonary artery simultaneously, along with slide tracheoplasty, in 60% with CPB assistance[5]. Previous preoperative ventilation is required, as in our case, in 31% of patients in the presence of stridor and respiratory distress in the neonatal resuscitation room. Difficulty in advancing a 3.5 mm ETT in a term newborn towards the distal airway should alert the pediatrician to the possibility of congenital airway obstruction. Immediately placing the ETT underneath the vocal cords or

a smaller one of 3 mm sufficient to ventilate the patient are acceptable measures to secure the airway and avoid acquired trauma added to intubation maneuvers that worsen the prognosis of CTS. Bronchoscopy is mandatory, as this approach will avoid the erroneous performance of a tracheostomy as an inadvertent procedure. The days of mechanical ventilation and the length of stay average 7 and 20 days, respectively. In-hospital mortality has been found to be related to the initiation of preoperative extracorporeal membrane oxygenation [ECMO] rather than the concomitant use of CPB. Prolonged length of stay is impacted by the complexity of the airway malformation and by systemic effects on the duration of CPB support, without impacting survival; the latter is affected by the need for simultaneous intracardiac surgery. Thus, isolated CTSs without pulmonary sling or without intracardiac malformations could benefit, as in our case, from slide tracheoplasty after 3 months of age without the risk factors associated with CPB: intraoperative hemorrhage with an increase in blood product transfusion, systemic effects of heparinization and fluid overload, the latter with an impact on the generation of edema and risk of dehiscence in the suture line of tracheal anastomosis.

Prolonged duration of postoperative mechanical ventilation has been associated with reinterventions in the airway, possibly favored by endotracheal trauma that adds local damage during endotracheal intubation strategies. Similarly, the complexity of CTS and age younger than 3 months at the time of slide tracheoplasty are other factors associated with in-hospital mortality[6].

Grillo et al.[7] published the first reports on slide tracheoplasty without the use of CPB, also corroborating satisfactory long-term airway growth after the intervention. Similar to our case, the original description by Tsang et al.[2] was modified by initially preparing with a medial and longitudinal incision on the anterior aspect of the distal tracheal portion an anterior slide that allows ventilation with a cuffed ETT appropriate for the patient's age of the airway and lungs during surgery, contrary to the original recommendation to incise the proximal segment anteriorly and the distal segment posteriorly under the support of CPB circulation. We started the anastomosis superiorly on the posterior wall, encountered no difficulties in completing the anastomosis, and there was no need to remove and reinsert the ETT to facilitate the anastomosis of the tracheal slide[7]. The other change was to spatulate the free edges of the preformed ends in such a way that a hermetic and perfect tongue-like overlap of both slides[2,8] was achieved, instead of the triangular arrangement of the original incisions that predisposed to a suture line overlapping the ends and fostering the characteristic "figure-eight" deformity on bronchoscopic view. We also believe that this modification avoids intraluminal cartilaginous exposure of said deformity as a predisposing factor for recurrent and refractory restenosis. The safety profile of this technique has been documented, sparing the use of CPB with bicaval cannulation with moderate hypothermia and cardioplegia to accompany intracardiac procedures or those with borderline cardiac physiology. In the absence of intracardiac repair, simple venous cannulation with normothermic CPB is sufficient. Current trends of early extubation from the immediate postoperative period and even non-invasive ventilation techniques (high-flow nasal ventilation, bilevel positive airway pressure [BiPAP], continuous positive airway pressure [CPAP]) are recommended, as they generate less local damage and edema on the anastomosis.

To optimize the most recommended surgical strategy for each patient based on mortality prognoses and associated cardiovascular anomalies, three groups have been proposed:

- With isolated long CTS, slide tracheoplasty could be recommended with or without the use of CPB support; low mortality group.
- With long CTS and non-complex vascular and cardiac anomalies (pulmonary artery sling, septal and atrial defects, etc.), slide tracheoplasty and the simple intracardiac anomaly could be corrected simultaneously.
- With long CTS and complicated cardiac and vascular anomalies (critical pulmonary stenosis, tetralogy of Fallot with absent pulmonary valve, right ventricular outflow tract obstruction, etc.), in which the duration of CPB is the main prognostic factor for mortality, a staged surgical strategy is suggested: initial airway stabilization (as in our case), slide tracheoplasty, and correction of the intracardiac heart disease at different surgical times.

In CTS with an extension of < 20% of the total tracheal length, tracheal resection and end-to-end anastomosis is recommended. In our case, it was possible to perform it as a reoperation for a local acquired lesion of stenosis due to intubation trauma and associated granulation tissue. It can be corroborated during reintervention tracheoplasty the transverse and

longitudinal tracheal growth, radiological and endoscopic, documented in several reports after slide tracheoplasty[9]. Detection and correction of major and minor complications should be performed from the immediate to late postoperative period. Major complications justify timely and early interventions; for example, anastomotic dehiscence within the first 10 postoperative days with signs of sepsis and air leak that will require immediate diagnostic bronchoscopy and stabilization of the airway with video-assisted endotracheal intubation, with temporary inflation of a balloon distal to the dehiscence, or selective bronchial intubation to improve conditions for an immediate and appropriate reoperation. Bronchoscopy, in all cases, as a minor intervention guarantees the potential success of conservative management with topical anti-inflammatory therapy and nebulized therapy with bronchodilation of some early sequelae associated with endotracheal intubation and mechanical ventilation from edema, evaluation of malacia and tracheobronchitis to early endoscopic resection of inflammatory granulomas. Minor complications, such as subacute or late formation of granulation tissue, may require, as in our case, removal of granulation tissue with forceps or pneumatic tracheal dilation with local and systemic steroids. In the presence of fibrotic recurrence and tracheal restenosis due to granulation tissue and refractoriness to previous endoscopic management, late and major interventions for stenotic resection and end-to-end tracheal anastomosis are justified, as occurred in the present case.

As in-hospital mortality prior to discharge and after slide tracheoplasty has dropped from 30% down to 6% due to multidisciplinary management with early recognition and treatment of complications, improvement in the variables of the surgical technique and intensive airway care, among others, late mortality after discharge remains stable at 10%, with recurrent airway restenosis being the main cause. In our case, recurrent and refractory restenosis due to granulation tissue formation was the main reason for reintervention (present in up to 23% of revisions, including tracheobronchial malacia). We have adopted the recommendation to recognize clinical signs of stridor and increased bronchial secretions and sequential bronchoscopic evaluation between extubation and 10 days, at 1, 3, and 6 months, and annually after slide tracheoplasty for the recognition and early intervention of these late complications[10].

Conclusions

Slide tracheoplasty is the surgical treatment of choice for complex CTS. Bronchoscopy is the ideal diagnostic and follow-up tool, along with coronary computed tomography angiography and evaluation of associated comorbidity. An acceptable surgical strategy can be offered; with a modification to the original description, it is possible, in our setting, to offer slide tracheal reconstruction without the risk factors associated with CPB and with the participation of a multidisciplinary team.

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Conflicts of interest

The authors declared no conflicts of interest.

Ethical considerations

Protection of persons and animals. The authors declare that no experiments on humans or animals have been performed for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed the confidentiality protocols of their institution, have obtained informed consent from the patients, and have the approval of the Ethics Committee. The recommendations of the SAGER guidelines have been followed, according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that they did not use any type of generative artificial intelligence for the writing of this manuscript.

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