Management of benign dynamic "A-shape" tracheal stenosis: a retrospective study of 60 patients

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Abstract

Benign tracheal stenosis complicates tracheal intubation or tracheostomy in 0.6% to 65% of cases. Surgical resection is the standard treatment. Endoscopic management is used for inoperable patients with 17% to 69% success. Dynamic "A-shape" tracheal stenosis (DATS) results in a dynamic stenosis with anterior fracture of tracheal cartilage and frequently associated posterior malacia. We report the results of our multidisciplinary management.

Reference


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Management of Benign Dynamic “A-Shape” Tracheal Stenosis: A Retrospective Study of 60 Patients

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**Background.** Benign tracheal stenosis complicates tracheal intubation or tracheostomy in 0.6% to 65% of cases. Surgical resection is the standard treatment. Endoscopic management is used for inoperable patients with 17% to 69% success. Dynamic “A-shape” tracheal stenosis (DATS) results in a dynamic stenosis with anterior fracture of tracheal cartilage and frequently associated posterior malacia. We report the results of our multidisciplinary management.

**Methods.** Sixty patients with DATS were included. Management decision was made during initial bronchoscopy. When suitable, patients were referred to thoracic surgery for tracheal resection. Posterior localized tracheomalacia was treated with laser photocoagulation of the posterior tracheal wall. Tracheal stents were placed if the stenosis persisted after laser treatment. The choice of stent (straight silicone, hour-glass shaped silicone, T-tube, or fully-covered self-expandable metallic stent) was based on operator’s judgment. After 12 to 18 months, stents were removed. If the stenosis persisted after stent removal, surgery was reconsidered. If surgery was not possible, a stent was replaced. In case of satisfactory result, a stent was replaced only after recurrence. Stable patients after treatment were considered as success, requirement of long-term tracheostomy or T tube as failure, and long-term stent as partial success.

**Results.** All patients developed DATS after tracheostomy. Thirty-three patients had posterior tracheomalacia. In 13 patients, mild stenosis required only endoscopic surveillance. Two patients were referred to thoracic surgery for tracheal resection surgery. Endoscopic management was the initial therapy in 45 patients (75%) and was considered successful in 23 patients (51%), partially successful in 10 (22%), and failed in 12 (27%). Five patients with successful outcomes required only laser therapy. Overall 70 stents were placed in 35 patients, with a migration rate of 31%.

**Conclusions.** The DATS management was successful in 63%. Stent migration was frequent. Posterior tracheomalacia was successfully treated in selected cases, avoiding stent placement.


Tracheal stenosis after tracheostomy can develop at the site of tracheostomy insertion, located in the upper third of the trachea, or at the cuff site, located in the middle or lower third of the trachea. Similar lesions are observed after tracheal intubation. Two major types of BTS are described in the classification proposed by Freitag and colleagues [11]. Structural stenoses, in which shrinking and scarring are predominant, are the most frequent. Dynamic stenoses are malacic conditions that vary with respiration. Another classification of BTS distinguishes simple web like stenosis and complex stenosis with cartilage damage, fibrous scarring with or without malacia [12]. Dynamic A-shape tracheal stenosis (DATS) is a particular type of structural or complex BTS that consists in dynamic stenosis due to anterior fracture of 1 cartilage ring often associated with posterior localized malacia.

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Despite structural stenosis, shrinking and scarring are seldom associated and the typical appearance is a triangular “A-shaped” or “tent-shaped” trachea (Figs 1, 2).

Surgical sleeve resection with end-to-end anastomosis is the standard treatment for BTS. The reported success rate ranges from 70% to 94% and the perioperative mortality is 1.5% to 9% [1, 13–17]. However, some patients are not considered candidates for surgery because of the extent of the stenosis (> 50% of the trachea or multifocal involvement) or because of any surgical contraindication. Endoscopic techniques have been developed as an alternative treatment [12, 18–26]. Dilatation, laser photo resection, and stent placement are the mainstay of endoscopic treatment strategy and afford excellent clinical results. An attempt to definitively remove the stent after 12 to 18 months is generally provided with variable long-term success (40% when the stent is placed for 6 months, 70% when it remains in place for 18 months) [12, 20–22]. Restenosis after stent removal is reported in up to 80%, requiring permanent stent placement or second line surgery [20]. The major drawback of stents is the possibility of migration, which happens in about 11% to 20% of all cases [12, 21, 26]. Even if very rarely life threatening, stent migration requires repeated intervention for stent removal and stenosis management.

Patients suffering from DATS are frequently excluded from studies describing endoscopic management of BTS [12, 20–22, 24]. As a matter of fact, DATS are not treatable with dilatation as there is no shrinking or scarring of trachea. Furthermore, because of the predominant dynamic component of this type of BTS, stent placement is generally thought to be complicated with a higher than usual migration rate. In this study, we report our management encompassing surgery and endoscopic treatments of patients suffering from DATS.

Patients and Methods

From January 1999 to July 2013, sixty consecutive patients referred for initial evaluation and management of post tracheostomy stenosis were diagnosed with benign DATS. Only patients with BTS reported to be of pseudoglottic type on the first bronchoscopy report were included in the present study. Other types of BTS were excluded. Demographic data were obtained (Table 1). All patients developed DATS after tracheostomy. Approval was obtained from the local institutional review board “Hôpital Nord ethical committee” for this retrospective chart study.

Management decision was made after the initial bronchoscopy, taking into account symptoms and temporary or definitive contraindication to surgery or general anesthesia. Endoscopic and surgical options were clearly explained to the patient prior to definitive management decision. Patients with surgical indication, who did not require temporary stent placement, were directly referred to surgeons. All endoscopic procedures were performed under general anesthesia with rigid bronchoscopy. Posterior localized malacia was treated with YAP (yttrium aluminum perovskite) laser (Fig 3). The YAP laser sessions could be repeated if necessary. Tracheal stents were placed in case of persistent stenosis after laser, as sole treatment or as a bridge to surgery (Figs 4, 5).

When a stent was required, the choice of stent was based on the operator’s best judgment, between straight silicone stent (Tracheobronxane TD, Novatech, La Ciotat, France) (Fig 4), stenotic hour-glass shaped silicone stent (Tracheobronxane ST, Novatech), Montgomery T tube stent (Novatech), or fully covered self-expandable metallic stent (SILMET, Novatech) (Fig 5). As described in previous studies, an attempt to remove the stent was done after 12 to 18 months. In case of a significant persistent stenosis, surgery was reconsidered. A stent was immediately replaced if surgery was deemed impossible. In case of adequate tracheal caliber, patients were followed and the stent was only replaced if a symptomatic tracheal stenosis recurred.

Stable patients after surgery or endoscopic treatment without any long-term tracheal material were considered as successfully treated. Requirement of long-term tracheostomy or Montgomery T stent was considered as failure. Requirement of long-term tracheal stent was considered as partial success, as these patients were subject to adverse effects and repeated stent replacement that can affect the quality of life [26].

Results

Of the 60 included patients for analysis, 53% (n = 32) were men. Mean age was 63 ± 13.7 years old. All patients developed DATS after tracheostomy. These patients underwent a total of 212 bronchoscopies (mean per patient = 3.5 ± 3.7). Mean duration of management from diagnosis to last therapeutic procedure was 407 ± 684 days with a median of 78 days. Length of follow-up after last therapeutic intervention was variable, with a mean of
542 ± 543 days and a median of 448 days. Thirty-three patients (55%) presented posterior tracheomalacia on the first evaluation bronchoscopy. The demographic data are summarized in Table 1.

Of the 60 patients, 13 (21.6%) had only mild stenosis with less than 30% reduction of the tracheal diameter. Follow-up confirmed that these patients did not develop worsening of their respiratory symptoms and did not require further treatment.

Two patients were referred for surgery after initial endoscopic evaluation. These 2 patients did well in the end but both had postoperative complications; suture dehiscence and hourglass like tracheal stenosis successfully treated with temporary stent.

Endoscopic treatment strategy was the first line of therapy in 45 (75%) patients. Five patients with successful management underwent only laser therapy for predominant posterior tracheomalacia. Fifteen successfully treated patients initially received a tracheal stent. During the course of management these patients required a mean of 2.2 stent (migration rate 33%) and 0.93 laser sessions. At the end of follow-up 13 patients were stabilized with a tracheal stent (Montgomery = 3, Silicone TD, ST, or metallic stent =10). Twenty patients were stabilized without stent and 7 patients required long-term tracheostomy.

A total of 70 stents were placed and distributed in 35 patients at some point in their management (range, 1 to 8 stents per patient). Sixteen (46%) patients received only 1 stent and 12 (34%) received 2 stents. The most frequently used was the TD stent (41 = 59%) with a mean diameter of 16.1 mm. Fourteen (20%) ST stents and 14 (20%) SILMET stents were inserted with a mean diameter of 15.86 mm and 18.46 mm, respectively. Only one Y stent was placed. Overall stent migration rate was 31% and occurred in 18 patients. Fifteen (83%) of them had only 1 stent migration episode. Stent migration rate was higher with ST stents (43%) and lower with SILMET stents (14%). Granuloma formation due to stent was reported in 5 (14%) patients, of whom 4 had received metallic stents.

Thirteen patients had successful stent removal attempts and 3 patients were operated after early stent migration without long-term recurrence of tracheal stenosis. Seven patients required long-term tracheostomy or Montgomery T tube for recurrence of stenosis after stent removal (n = 2) or early stent migration (n = 5) and surgery contraindication. Two patients suffered from recurrence of stenosis after stent removal, 1 of whom was considered as a surgical candidate but was lost to follow-up, and the other was an intensive care unit patient with contraindication to surgery and deceased from an unrelated cause. These 2 patients were considered as management failure. Two patients died from unrelated causes.

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Table 1. Demographic and Management Strategy of Study Population

<table>
<thead>
<tr>
<th>Variable</th>
<th>Population (n = 60)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Demography</td>
<td></td>
</tr>
<tr>
<td>Age – year ± SD</td>
<td>63 ± 13.7</td>
</tr>
<tr>
<td>Sex ratio: nb (%)</td>
<td>32 (53) M/28 (47) F</td>
</tr>
<tr>
<td>Localized posterior malacia: nb %</td>
<td>33 (55)</td>
</tr>
<tr>
<td>Initial management strategy</td>
<td></td>
</tr>
<tr>
<td>Surgery: nb (%)</td>
<td>2 (3)</td>
</tr>
<tr>
<td>Endoscopic: nb (%)</td>
<td>45 (75)</td>
</tr>
<tr>
<td>No treatment required: nb (%)</td>
<td>13 (22)</td>
</tr>
<tr>
<td>Endoscopic management: first procedure</td>
<td></td>
</tr>
<tr>
<td>Stent placement: nb (%)</td>
<td>38 (85)</td>
</tr>
<tr>
<td>Laser therapy: nb (%)</td>
<td>6 (13)</td>
</tr>
<tr>
<td>Tracheostomy: nb (%)</td>
<td>1 (2)</td>
</tr>
<tr>
<td>Tracheal stents</td>
<td></td>
</tr>
<tr>
<td>Overall: nb (migration rate %)</td>
<td>70 (31)</td>
</tr>
<tr>
<td>TD stent: nb (%/migration rate %)</td>
<td>41 (59/34)</td>
</tr>
<tr>
<td>ST stent: nb (%/migration rate %)</td>
<td>14 (20/43)</td>
</tr>
<tr>
<td>SEMS stent: nb (%/migration rate %)</td>
<td>14 (20/14)</td>
</tr>
</tbody>
</table>

nb = number; SEMS = self expanding metallic stent; ST = hourglass silicone stent; TD = straight silicone stent.
before 18-month stent removal attempt, and 8 patients were lost to follow-up before stent removal could be attempted. These last 10 patients were considered as partial success of management.

Overall, endoscopic management strategy proved successful in 23 patients (51%), partially successful in 10 patients (22%), and failed in 12 patients (27%) (Fig 6). Overall, management succeeded in 38 patients (63%).

Comment

Tracheal stenosis is a well-described complication of intubation or tracheostomy. Endoscopic management successfully treats simple tracheal stenosis in most cases [23]. Surgery is the reference treatment but cannot be provided in some patients, fails in up to 15% of cases, and the perioperative mortality rate is up to 9%. For these reasons, endoscopic treatment based on dilatation and stent placement was developed, either as definitive therapy or as bridge to surgery.

Dynamic A-shape tracheal stenosis is not amenable to dilatation. In addition, because of the predominant dynamic component of the stenosis and the absence of concentric scarring, stent migration is frequent. For these reasons, DATS are frequently excluded from endoscopic management studies [12, 20–22, 24].

To our knowledge, this is the first description of management of patients suffering from DATS. Endoscopic procedure was the first line of therapy in the majority of patients, with a success rate of 51% without the need for further intervention, and in another 22% in whom long-lasting tracheal stents were required. As expected, stent migration rate was high (31%) and is certainly related to the anatomic characteristics of DATS compared with other types of BTS. This can be reduced by external fixation [27, 28]. Interestingly, in our study migration tended to be lower with the use of metallic stents, which could be attributed to their higher mean diameter and their better adaptation to the shape and dynamic component of DATS. The use of a dedicated hour-glass silicone stent (ST stent) showed a very low migration rate in other types of BTS. However, for DATS, migration rate of this stent was the highest, most probably due to their lower mean diameter, as the higher commercialized diameter of ST stents at the time of this study was 16 mm compared with 20 mm for metallic stents [29]. On the other hand, metallic stents tended to induce more granulation tissue. Laser therapy in the treatment of airway collapse is a new therapeutical approach [30].
Airway stability relies on the ratio between the length of the cartilaginous portion (C) and of the posterior membrane (M). A normal ratio is approximately 4:1. In tracheobronchomalacia it can reach 2:1 due to a reduced amount of cartilage or an excess of posterior membrane length [31]. In the case of DATS, the damaged cartilaginous ring induces a decrease in its length and an increase of the C/M ratio without direct damage to the posterior membrane. To restore the C/M ratio and the stability of the airway segment, laser therapy aims to shrink the posterior localized expiratory collapse (Fig 3); it was an alternative to stent placement for 5 well-selected patients who suffered from moderate stenosis with predominant posterior localized malacia. The 100% success rate in these patients confirms that laser therapy is promising and should be considered for these selected patients.

Our study has some limitations that need to be mentioned. It is a retrospective monocentric cohort study and, as such was not designed to demonstrate different outcomes from statistical and power perspectives. The numbers in some of the sub-cohorts are limited and the results should be interpreted with caution. Some of our patients were lost to follow-up and could not be evaluated for long-term success. In particular, an attempt to remove the tracheal stent could not be provided after 12 to 18 months in some of them. The endoscopic management strategy was the first-line strategy in the majority of our patients and very few patients required surgery. Patients’ comorbidities and emergency at initial presentation partly explain this management strategy, but there is a center-effect favoring endoscopic management. In fact, there was no absolute contraindication to surgery in all of...
our patients. Contraindication to surgery is an inadequate terminology because it relies on definitive or temporary anesthetic contraindications. For some of our patients, surgery could have been postponed while waiting for a global health status improvement. In a more general note, tracheal stenosis management in our institution is somehow biased, given that there was an excessive tendency to use stents as the primary treatment for this condition from 1999 to 2008. Even though we state that surgery is the gold standard treatment for tracheal stenosis, we used to favor the endoscopic management as the first step and to consider surgery only after failure of stenting. Our practice has evolved but this does not appear clearly in this study, which covers a period of 14 years. Since 2008 we explain to our patients both options (surgery and endoscopy), insisting on the necessity of surgery. A large number of patients still prefer a noninvasive approach as first step management and surgery in case of failure. Figure 7 proposes an algorithm for management of these patients.

In all of our patients considered as failure, who were not lost to follow-up, surgery could not be done because of anesthetic contraindications. For all of the patients considered as partial success (long-term stenting) who were not lost to follow-up, surgery was continuously refused by the patients. They deemed stenting a satisfactory management despite daily maintenance with nebulization of saline and stent annual replacement, which is necessary to estimate the need to reinsert a new one. For all these reasons, our results should not be over generalized.

In summary, the results of this single-center experience on DATS multidisciplinary management strategy add to a missing literature in this specific field. Our results confirm that multidisciplinary management is safe and highly effective for the majority of patients. They also confirm that stent migration rate is higher than in other BTS types, but could be lower for self-expanding metallic stents. Finally, laser therapy of posterior localized malacia provided excellent results in selected cases and avoided surgery or stent placement. Prospective studies should be provided for confirmation and generalization of these results.

References

INVITED COMMENTARY

Plojoux and colleagues [1] present their experience with primary endoscopic management of 60 patients with posttracheostomy tracheal stenosis. This particular lesion has been well described as a stomal complication of tracheostomy, with several factors felt to contribute to this complication. Oversizing of the tracheostomy tube to the size of the patient’s trachea, creation of too large an anterior tracheal defect at the time of tube placement, and leveraging against the wall of the stoma from unsupported ventilator tubing are important etiologic factors. The trachea is inevitably contaminated with bacteria and this too may play a role in the erosion of cartilage and the development of the A-shaped stenosis. Grillo [2] has quite clearly described the etiology and treatment of the condition. Figure 1 demonstrates the pathogenesis of this phenomenon.

This report is interesting because it presents a multifaceted endoscopic approach with laser photoablation and stenting for the management of what in most centers is a surgical problem. The authors recognize and accept this. They also recognize that their contraindications for surgical intervention were often temporal in nature and that surgical intervention could have been done at a later time. Of the 60 patients, 13 required no therapy. Of the 47 remaining patients, 5 eventually had surgical resection, 20 had successful stabilization without a stent, 7 patients required a long-term tracheostomy, and 3 required a long-term T tube. Of the 13 patients treated with a long-term stent, 10 required repeated long-term stents and were considered “partial successes.” To summarize, the treatment successes were 5 surgical patients, 20 of 22 patients who were managed without a stent, and the partial success of 10 long-term stent patients.

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