CASE REPORT

Thoracic oesophageal cancer as a cause of stridor: a literature review

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SUMMARY

An 83-year-old woman, with a background of treated squamous cell oesophageal cancer, presented with a 3-week history of stridor. Of note, the patient had no risk factors for oesophageal cancer other than age. Clinical examination was unremarkable apart from stridor. Laboratory investigations, including arterial blood gas on room air, were unremarkable. Radiological examination revealed a 4.5×3.5×3.6 cm mass involving the posterior trachea and invading the tracheal orifice. Oesophagogastroduodenoscopy and rigid bronchoscopy confirmed an extensive tumour arising from the lower oesophagus and invading the trachea, causing 90% airway obstruction for a 6 mm length ending 1.5 cm above the carina. Biopsy revealed a poorly differentiated carcinoma with foci of squamous cell carcinoma. Unfortunately, the patient passed away 2 months after palliative tracheal stent placement.

BACKGROUND

Oesophageal cancers are primary or metastatic malignant neoplasms in the oesophagus.¹ They constitute 11% of all mediastinal masses² and are one of the deadliest forms, with a 5-year survival rate of 40%.³

Patients typically present late, with the majority of symptoms occurring once the neoplasm becomes invasive.⁴ They can therefore present with a range of symptoms. An evaluative study by the American College of Surgeons reported dysphagia (74%), weight loss (57.3%), reflux (20.5%), odynophagia (16.6%), dyspnoea (12.1%) and chronic cough to be the most common presenting symptoms.⁵ Rarely, however, has an airway emergency been the initial and only presentation.

CASE PRESENTATION

An 83-year-old woman, with a background of squamous cell oesophageal cancer, presented with a 3-week history of stridor and dyspnoea.

She was initially diagnosed with early stage oesophageal cancer 3 years prior to this presentation and had received treatment in the form of endoscopic mucosal resection surgery and adjuvant chemotherapy before being discharged from follow-up after a period of clear surveillance. Unfortunately, full immunohistochemical staining results, chemotherapy regimen details and details on interval of surveillance were not available as she had been treated in another region. Of note,

however, the patient was Caucasian, had never smoked, was not obese, had no history of high alcohol intake or, indeed, of Barrett's oesophagus. She also had no history of respiratory or cardiac disease. She denied upper aerodigestive symptoms, including those listed by the American College of Surgeons.⁵ She had always been self-caring at home.

Clinical examination was unremarkable apart from stridor. Laboratory investigations, including arterial blood gas on room air, were unremarkable.

Radiological examination by chest radiography (CXR) and CT neck, thorax, abdomen and pelvis revealed a 4.5×3.5×3.6 cm mass involving the posterior trachea and invading the tracheal orifice (figures 1–3), with no metastases and were otherwise unremarkable. Subsequent rigid bronchoscopy and fiberoptic oesophagogastroduodenoscopy (OGD) confirmed an extensive oesophageal tumour arising from the lower oesophagus and invading the trachea, causing 90% airway obstruction for a 6 mm length ending 1.5 cm above the carina. Biopsy revealed a poorly differentiated carcinoma with foci of squamous cell carcinoma. There was no tracheo-oesophageal fistula found at these examinations.

The patient was treated with palliative tracheal stent placement after Multi Disciplinary Team discussion (figure 4). Unfortunately, she passed away 2 months later.

INVESTIGATIONS

Chest radiograph.

Figure 1 Posterior Anterior (PA) CXR on admission.

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CT neck, thorax, abdomen and pelvis.
Rigid bronchoscopy.
Fibreoptic endoscopy.

TREATMENT

Palliative tracheal stent placement.

OUTCOME AND FOLLOW-UP

She passed away 2 months later.

DISCUSSION

With such a rare presentation and no randomised controlled trials available to inform management and prognostic advice, a review of the literature was undertaken to make explicit the frequency of this phenomenon and the management options available under these circumstances.

Literature review

Methodology

Relevant literature regarding stridor as a consequence of oesophageal cancer was identified through the use of biomedical databases for primary research material. The purpose of this review was to establish the incidence of this 'rare' occurrence, the demographics of those presenting with it, the short-term and long-term management as well as the overall prognosis. Two universal research databases were therefore used to identify publications from inception until present (February 2017): Medline and Embase. Key publications were further identified from Google Scholar and citations of publications analysed during this review.

Search terms were broadened as much as possible to develop a comprehensive list of results. 'Malignancy', 'cancer', 'tumour' and 'neoplasm' were used synonymously while 'oesophageal' and 'esophageal' were interchanged to address the American counterparts. Truncation symbols were used to further extend the search. 'Stridor' was a key term for inclusion and so used alone.

A range of literature was gathered, including journal articles and conference abstracts. These were manually searched for

Figure 3 CT coronal on admission.

relevant material by the two authors (RMM and VM). Duplicates, irrelevant and non-English citations were excluded. The resultant list of publications was further reviewed to determine eligibility. This was dependent on whether the reported stridor was caused directly by an oesophageal carcinoma, either through invasion or compression of the airway. Stridor as a consequence of management was excluded. The final shortlist of articles⁶⁻¹⁹ was agreed by consensus among both authors.

Results

The database search yielded 51 citations with 8 identified through screening citations from Google Scholar. Once duplicates were removed, 44 records remained for screening. Each article was reviewed by both authors, with 31 removed in consensus for not meeting the set criteria, that is, not relating to cases of stridor due to oesophageal cancer. The remaining articles were analysed, and the results summarised in tables 1-4. These were all case reports or case series (table 1). Significantly, there were no cohort studies to extract incidence rates, nor were there any randomised controlled trials.

Figure 2 CT axial on admission.

Figure 4 Poststent placement.

Table 1 Summary of included publication characteristics

	All
Types of publications	
Case report	8
Case series	5
Cross-sectional study	0
RCT*	0
Systematic review	0
Type of article	
Journal article	10
Conference abstract	2
Journal letter	1

*Randomised controlled trial.

Overall, there were 26 patients documented (table 2), with a mean age of 60, most of whom were men. Most of the patients were known to have a history of oesophageal cancer, with surgery and radiochemotherapy being the most popular treatment modalities used in these patients.

Table 3 summarises the signs and symptoms observed at presentation. Dyspnoea was the most common symptom after stridor, which was present in all patients. This was followed by dysphagia. Eight patients had no other clinical features reported; however, sole stridor was not exclusively mentioned.

Tracheal stenosis/invasion was found to be the cause of stridor in 25 of the 26 patients, either alone or associated with vocal cord paralysis, which was responsible for stridor in seven patients. Four individuals were found to have tracheoesophageal fistulas. Squamous cell carcinomas were the most frequent tissue type of the causative carcinoma, although an adenocarcinoma occurred in one of the cases.

Table 4 summarises the management of the 26 patients. Tracheobronchial stenting was the most documented

Table 3 Summary of patient evaluation

	All
Total	26
Presenting symptoms*†	
Dyspnoea‡	18
Tachypnoea	3
Dysphagia	5
Dysphonia	1
Loss of weight	2
Lymphadenopathy	2
Cough	3
None	6
Not stated	1
Cause*	
Tracheal invasion/stenosis	25
Vocal cord paralysis	7
Fistula	4
Histology	
Squamous cell carcinoma	11
Adenocarcinoma	1
Not stated	14
Percentage of stenosis	
>90	2
70–89	1
50–69	1
20–49	1
1–19	0
Not stated	24

*Numbers do not equal total as patient may have had more than one symptom/cause.

†Excludes stridor as all patients had stridor.

‡Includes 'respiratory distress' and 'shortness of breath'.

management strategy, used in 22 of the cases—13 of these were inserted acutely and left as the definitive management, 3 were inserted acutely to achieve stability and 6 were inserted definitively following other acute methods. In the cases reporting progress postmanagement, the mean survival time was 2.45 months with one individual surviving beyond follow-up (2 months).

Surgery was used alone or in conjunction with other treatment modalities in nine patients. Cordotomy was used in six patients, all acutely, and tracheostomy in two. Cricothyroidotomy was performed on one individual. Mean survival postcordotomy was 1.875 months, with one patient lost to follow-up and another living beyond the follow-up period of 2 months. Cordotomy was carried out with stent placement in the four remaining patients.

Of the four patients not stented, one was lost to follow, two did not have any follow-up period stated and the remaining one patient survived for 3 weeks.

Medical management, which included steroids, bronchodilators, oxygen and high-frequency jet ventilation, was acutely employed in only three patients. Radiotherapy (3), intubation (4), extracorporeal membranous oxygenation (2) and chemotherapy (1) were also used.

DISCUSSION

Stridor is widely recognised as an airway emergency, indicating significant extrathoracic or intrathoracic airway obstruction. It is associated with children more than adults due to their smaller airways and underdeveloped accessory muscles, thus differing in aetiology and management.²⁰ While common causes among children include croup, epiglottitis and foreign bodies, adults

Table 2 Summary of patient characteristics

	All
Total	26
Age	
Mean	60
Max	78
Min	43
Not stated	5
Gender	
Male	11
Female	4
Not stated	11
Oesophageal cancer	
New diagnosis	4
Known history	13
Not stated	9
Previous treatment*	
Surgery	7
Chemotherapy	1
Radiotherapy	3
Brachytherapy	3
Radiochemotherapy	6
No treatment	1
Not stated	5

*Numbers do not equal total as patient may have received more than one treatment.

Table 4 Summary of management

	All
Total	26
Stenting	
Acute and definitive	13
Definitive only	6
Acute only	3
Total	22
Surgery	
Acute tracheostomy	1
Delayed tracheostomy	1
Acute cordotomy	8
Delayed cordotomy	3
Delayed cricothyroidotomy	1
Other	
Medical*—acute	3
Intubation—acute	3
Radiotherapy—delayed	3
Extracorporeal membrane oxygenation—acute	1
Cardiopulmonary bypass—acute	1
Chemotherapy—delayed	1
Survival poststenting	In months
Survival mean†	2.45
Lost to follow-up	Two patients
Surviving	One patient
Not stated	Eight patients

*Includes steroids, bronchodilators, oxygen and high-frequency jet ventilation.

†In months.

encounter stridor as a result of neurological causes, vocal cord diseases and local lesions.^{20 21} Mediastinal masses fall under this final category, 11% of which are of oesophageal origin.²

Other oesophageal causes of stridor have been noted, including gastro-oesophageal reflux disease, foreign bodies in the oesophagus, oesophageal dysmotility and incoordination between the upper oesophageal sphincter and the rest of the alimentary tract, with accompanying oesophageal dilation suspected to cause extrinsic compression of the trachea in the latter cases and gastro-oesophageal reflux being a factor in paediatric rather than adult stridor.^{22–25}

It is interesting to note that this patient did not have any risk factors for oesophageal cancer other than age, though diet may have played a factor; however, it is difficult to trace back her diet over such a long life span.⁵ The fact that both the primary and the new malignancy are morphologically squamous cell based suggests that this may be a recurrence; however, it is possible that this is a new primary malignancy as patients with aerodigestive tumours are at increased risk of multiple primary cancers in the aerodigestive tract. And, second primary tumours, even in the absence of previous mediastinal radiation, as with our patient, may occur—with a stated incidence of 0.9%, with field carcinogenesis sometimes being implicated in this phenomenon.^{26–28} Unfortunately, due to the patient being treated in another region, it was not possible to ascertain the specific immunohistochemistry results, chemotherapy regimen given or the imaging and interval of surveillance, which makes it difficult to have a clear idea of what the prognosis was at initial presentation, as well as to be clearer on whether or not this is due to progression or a second primary malignancy.

Furthermore, it was important to consider the possibility of tracheoesophageal fistulation, as this can be a rare but

life-threatening complication of oesophageal surgery presenting years later,²⁹ and there is a suggestion of mediastinal air in the CT scans which would indicate the presence of one; however, there was no indication of this at bronchoscopy or OGD.

This literature review exclusively identified case series/reports, suggesting that the researched phenomenon has not been sufficiently common to warrant cohort studies. McGrath *et al*'s case series did identify five patients with stridor over an 8-year period; however, the study focused on tracheoesophageal stent placement in that period of time rather than patients presenting with stridor, casting this incidence rate into doubt.

The average age of patients presenting with this origin of stridor was found to be younger than the 65–70 years old cited in the literature.³⁰ This suggests a more aggressive progression of this carcinoma in these patients, commencing earlier than in their non-stridulous counterparts. The review further supports this, with a large proportion of patients having a background of treated oesophageal carcinoma.

As an airway emergency, stridor should be managed according to the adult life support principles. It is therefore significant that only three papers discussed medical management, which we believe to be consequence of under-reporting. It is a failure of this approach which then necessitates further acute or definitive intervention, the most reported of which were stent placement and surgery.

Tracheobronchial stenting was most commonly used, both in the acute and delayed stages. The majority cases of stridor were brought on by tracheal stenosis, due to infiltration or compression, with even 20% stenosis being sufficient to elicit the sign. The direct tamponade of a stent therefore renders it successful in immediate symptomatic relief. Su *et al*'s study in 2004 retrospectively analysed 10 patients with respiratory distress due to obstruction, who were treated with stent placement between 1999 and 2003; 9 out of the 10 individuals experienced immediate improvement post procedure.³¹ Our review identified successful outcomes in 20 out of the 22. This 91% success rate was in keeping with the 82%–97% range identified by Bacon *et al*'s review of outcomes in metal stent use in tracheal stenosis.³²

The long-term outcomes, however, seem to favour its role in palliation, with patients enduring a mean survival of 2.45 months. In fact, an audit reported by Conacher's review article highlighted that two-thirds of deaths poststent placement were within 10 weeks of the procedure, due to its palliative use in terminal cancers.³³ This role is further supported by Lemaire *et al*'s³⁴ retrospective study in 2005 analysing stent placement in 140 patients, which came to the conclusion that there was an increased complication rate associated with stent situation for >30 days.²⁶ Complications were reported in five of our reviewed cases—four patients required revision due to stent migration (2) and other causes (1) and one suffered odynophagia postoperatively—however, the duration of stent placement had not been reported.

Significantly, stenting can be used in conjunction with other interventions, including surgery and radiotherapy. It can maintain airway patency following debridement or removal of surrounding structures, or more frequently be an avenue to ensure better definitive outcomes of these procedures.³² Stenting combined with surgery and radiotherapy ensured successful outcomes in 6 out of 8 patients.

Surgical procedures were performed in nine cases, including cordotomy (6), tracheostomy (2) and cricothyroidotomy (1). Failure of endotracheal therapies usually precede these procedures.³⁴ The latter two are indicated when there is involvement above the level of the vocal cords and performed to achieve

airway control, alleviating symptoms in 2 out of 3 cases.³⁵ Vocal cord paralysis, which was the second major mechanism eliciting stridor, is conventionally managed with a cordotomy to reform the glottis and was further supplemented with stenting.

Treatments such as extracorporeal membrane oxygenation and cardiopulmonary bypass were used acutely, a common bridging practice to ensure stability before definitive management, that is, stent placement. Willms *et al*'s literature review found 12 cases of extracorporeal membrane oxygenation use in emergencies pertaining to acute tracheal obstruction between 1989 and 2010; in all cases, the patient survived to hospital discharge following definitive intervention.³⁶ Similar success was seen in the two cases found in our review.

Conclusion

Our patient presented with acute stridor due to tumour infiltration at the lower segment of the trachea, below the level of the vocal cords. She had a history of oesophageal cancer, fitting the demographic identified in the review. No additional medical relief or bridging treatments were required given the stability of the patient and given the location of the lesion; tracheobronchial stenting seemed the most appropriate intervention in the acute setting.

The variety of treatments identified by our review show patients should be treated in a bespoke manner and according to the resources available, with patients managed using adult life support principles, and being suitably escalated and potentially transferred to specialist centres to ensure adequate oxygenation and specific definitive management.

Learning points

- Stridor should be managed according to adult life support principles.
- Stridor as a result of oesophageal cancer portends a poor outcome.
- Tracheobronchial stenting is the most commonly used treatment for stridor as a result of oesophageal cancer.
- Definitive treatment of stridor as a result of oesophageal cancer must be based on cause.
- Symptomatic improvement is achievable, but prognosis is ultimately poor regardless of intervention.

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