Long-segment tracheal stenosis: slide tracheoplasty and a multidisciplinary approach improve outcomes and reduce costs

Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

Health technology
A multidisciplinary approach for the management of long-segment tracheal stenosis (LSTS) in children was examined. The multidisciplinary tracheal team (MDTT) comprised the following aspects

- all referrals of children with tracheal problems would be channelled through the MDTT;
- investigations would be carried out according to a fixed protocol;
- slide tracheoplasty would become the procedure of first choice and would be extended to include longer-segment stenosis;
- coexistent congenital heart defects would be fixed at the same operation;
- patch tracheoplasty would become a second-string strategy;
- tracheal homograft repair would be used for recurrent severe stenosis if stenting failed;
- postoperative surveillance would be by fibre-optic bronchoscopy and bronchography;
- granulations would be managed with radial balloon dilatation;
- members of the team would be cross-skilled to facilitate timely follow-up;
- nurse liaison and lead administrator posts would be established to improve integration of care pathways and communication with patients, families, referrers and the multiple teams involved;
- shared care with referring units would be encouraged and facilitated;
- a weekly meeting of the MDTT would be held for patient and data review.

Type of intervention
Treatment.

Economic study type
Cost-effectiveness analysis.

Study population
The study population comprised patients aged less than 16 years with a primary diagnosis of LSTS for more than two thirds of the length of the trachea and involving the carina.
Setting
The setting was a hospital. The economic study was carried out in the UK.

Dates to which data relate
The effectiveness and resource use data were gathered from 1998 to 2000 for the pre-MDTT period, and from 2001 to 2004 for the post-MDTT period. The price year was not reported.

Source of effectiveness data
The effectiveness evidence was derived from a single study.

Link between effectiveness and cost data
The costing was performed retrospectively on the same sample of patients as that used in the effectiveness study.

Study sample
Power calculations were not reported. A sample of 34 patients was identified and included in the study. There were 19 patients (13 boys) in the pre-MDTT group and 15 patients (11 boys) in the post-MDTT group. The mean age of the patients was 18.7 weeks (age range: 1.3 - 104.2) in the pre-MDTT group and 14.42 weeks (age range: 1.8 to 782.1) in the post-MDTT group. Further details of the methods of sample selection were not reported.

Study design
This was a retrospective comparative study with historical control that was carried out at a single centre. Patients who survived after hospital discharge were followed for an average of 38 months (range: 2 - 57). No patient was lost to the follow-up assessment.

Analysis of effectiveness
All of the patients included in the initial study sample were accounted for in the analysis of effectiveness. The outcome measures were:

the mortality associated with the two treatment approaches,

the length of stay (LOS) in the intensive care unit (ICU),

the duration of endotracheal intubation,

the LOS in the high-dependency unit (HDU), and

the total LOS in the hospital.

The baseline comparability of the study groups was not discussed.

Effectiveness results
In the pre-MDTT period, 2 (17%) of the 12 patients who had a suspended pericardial patch tracheoplasty died, 2 (67%) of the 3 patients who had a simple unsuspended patch died, and 2 (50%) of the 4 patients who underwent repair by means of a tracheal autograft technique died. Two patients in both groups died during the follow-up period.

In the post-MDTT period, only 2 (13%) of the 15 patients who underwent SPT died.

The LOS in the ICU was 28 days (range: 5 - 130) in the pre-MDTT period and 7 days (range: 2 - 54) in the post-MDTT period, (p=0.03).
The duration of endotracheal intubation was 17.8 days (range: 0.7 - 40.4) in the pre-MDTT period and 5.2 days (range: 1.7 - 54) in the post-MDTT period, (p=0.09).

The LOS in the HDU was 17 days (range: 1 - 130) in the pre-MDTT period and 5 days (range: 1 - 26) in the post-MDTT period, (p=0.78).

The total LOS in the hospital was 59 days (range: 5 - 243) in the pre-MDTT period and 21.5 days (range: 10.4 - 55) in the post-MDTT period, (p=0.01).

Clinical conclusions
The effectiveness analysis showed that the MDTT approach led to lower mortality and shorter hospital stay.

Measure of benefits used in the economic analysis
The health outcomes were left disaggregated and no summary benefit measure was used in the economic analysis. In effect, a cost-consequences analysis was carried out.

Direct costs
Discounting was not relevant since the costs were incurred during a short timeframe. The unit costs were not presented separately from the quantities of resources used. The economic evaluation considered costs associated with LOS in the ICU and HDU, as well as the costs of treatment. The cost/resource boundary of the study was not reported. The source of the data was not given. Resource use was estimated from the same sample of patients as that used to derive the clinical data. The price year was not reported.

Statistical analysis of costs
An unpaired t-test was used to test the statistical significance of differences in the estimated costs.

Indirect Costs
The indirect costs were not included in the economic evaluation.

Currency
UK pounds sterling (€).

Sensitivity analysis
No sensitivity analyses were performed.

Estimated benefits used in the economic analysis
See the 'Effectiveness Results' section.

Cost results
The median ICU costs per patient were 41,208 (range: 8,585 - 223,210) in the pre-MDTT period and 12,019 (range: 6,868 - 92,718) in the post-MDTT period, (p=0.03).

The median HDU costs per patient were 13,209 (range: 777 - 101,010) in the pre-MDTT period and 3,885 (range: 777 - 20,202) in the post-MDTT period, (p=0.07).

The median total costs per patient were 56,331 (range: 8,585 - 324,220) in the pre-MDTT period and 18,401 (range:
13,648 - 92,718) in the post-MDTT period, (p=0.01).

The cost of treatment per patient was 77,333 in the pre-MDTT period and 26,219 in the post-MDTT period (difference 51,114).

**Synthesis of costs and benefits**
A synthesis of the costs and benefits was not relevant since a cost-consequences analysis was performed.

**Authors’ conclusions**
Children with long-segment tracheal stenosis (LSTS) could benefit from a multidisciplinary team approach and a policy of primary slide tracheoplasty.

**CRD COMMENTARY - Selection of comparators**
The selection of the comparator was appropriate as it reflected the standard approach before the introduction of the MDTT. Both comparators were clearly described. You should decide whether they are valid comparators in your own setting.

**Validity of estimate of measure of effectiveness**
The effectiveness evidence came from a retrospective comparative study, which generally suffers from several limitations. For example, the fact that the two study groups were not studied simultaneously and factors other than the study intervention could have affected the results of the analysis. Further, a small sample of patients was identified and there was no evidence about the appropriateness of this sample. Similarly, it was unclear whether the two groups of patients were comparable at baseline. In addition, there was limited information on the methods used to assess the outcomes. In fact, the authors noted that the design of the study precluded a formal comparison of strategies. The authors also pointed out that the effect of era and learning curve might have accounted for the study findings.

**Validity of estimate of measure of benefit**
No summary benefit measure was used in the analysis because a cost-consequences analysis was conducted. Please refer to the comments in the "Validity of estimate of measure of effectiveness" field (above).

**Validity of estimate of costs**
The authors did not state explicitly the perspective of the study and only those costs related to the duration of hospitalisation were included in the analysis. The source of the costs was not reported. Similarly, the unit costs were not provided and this limits the replication of the analysis. Statistical tests were performed to examine the statistical significance of differences in the costs. The price year was not reported, which reduces the possibility of refleting the results of the analysis to other settings. The cost estimates were specific to the study setting.

**Other issues**
The authors did not make extensive comparisons of their findings with those from other studies. They also did not explicitly address the issue of the generalisability of the study results to other settings. Sensitivity analyses were not carried out, which further reduces the external validity of the analysis. The authors noted some limitations to the validity of their study, which have been reported already.

**Implications of the study**
The study results supported the use of a multidisciplinary team approach and slide tracheoplasty for the management of children with LSTS. The authors noted that, ideally, well-designed prospective studies should be carried out to examine...
the clinical and economic impact of the new multidisciplinary approach. However, it was stressed that the available patient population for such studies is quite small.

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