Role of skin-lined tracheotomy in obstructive sleep apnoea syndrome: personal experience

La tracheotomia “skin-lined” nel trattamento dei disordini ostruttivi respiratori del sonno: nostra esperienza

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Summary

Permanent tracheotomy was the first surgical procedure proposed for the treatment of severe obstructive sleep apnoea syndrome and is still the only surgical option that ensures, even in very severe cases, complete elimination of apnoea and, in turn, clinical remission. Improved knowledge of the causes of obstructive sleep apnoea syndromes and the increasing therapeutic options (instrumental, medical and surgical) have resulted in cases requiring tracheotomy as the only indispensable therapeutic option becoming more rare. At present, the only indications are in very occasional conditions of life-threatening obstructive sleep apnoea syndromes and in patients with continuous positive airway pressure is not tolerated or is not effective (severe deoxygenation or hypercapnia, severe respiratory disorder index, severe obstructive sleep apnoea syndrome-related arrhythmias, severe excessive daytime sleepiness, heart diseases or ischaemic encephalopathy exacerbated by obstructive sleep apnoea syndromes, obstructive pneumopathy exacerbated by obstructive sleep apnoea syndromes, severe obstructive sleep apnoea syndromes with few chances of resolution with other surgical procedures or failure of the latter). Moreover, it is the only therapeutic solution in rare nocturnal laryngeal stridor due to multisystemic atrophy (in which obstructive sleep apnoea syndrome is due to nocturnal laryngospasm of neurologic origin). Therapeutic tracheotomy must be permanent (tracheotomy) and, therefore, preferably carried out with a specific technique (skin-lined tracheotomy), able to guarantee greater stability, less risk of granulation tissue, wider opening of the tracheostomy, sufficient reversibility. In our experience, very few patients (10 cases) with sleep disorder breathing have been submitted to skin-lined tracheotomy. Of these, the majority were submitted to surgery for severe apnoea due to nocturnal laryngospasm on account of multisystemic atrophy (n = 7), while only 3 cases of obstructive sleep apnoea syndromes were submitted to skin-lined tracheotomy, i.e., 0.7% of the 424 patients operated on for obstructive sleep apnoea syndrome and 1.7% of the 175 operated on for severe, or very severe, obstructive sleep apnoea syndromes (RDI >40). Skin-lined tracheotomy was not followed by important complications and expected results were achieved with immediate disappearance of daytime symptoms and considerable improvement in nocturnal apnoea. Besides sleep-related disorders, numerous clinical situations with indications for a permanent tracheotomy may benefit from the skin-lined technique, such as severe laryngeal or tracheal stenoses, laryngeal diplegias, miasthenia gravis, lateral amyotrophic sclerosis, intractable aspiration, severe emphysema.
Introduction

In the Fifties, the obstructive sleep apnoea syndrome (OSAS) had not yet been identified from a pathogenetic and physiopathological point of view and the clinical pattern, characterised by pathological daytime sleepiness and by cardio- and cerebro-vascular complications, was erroneously correlated with a status of chronic alveolar hypoventilation, related to the grade of obesity of the patients studied (so-called pickwickian syndrome) 1. In the Sixties, the first polysomnographic recordings demonstrating that the pathogenesis of the daytime sleepiness typical of pickwickian syndrome was due to repeated episodes of prolonged obstruction at upper airways and not to a status of chronic carbonarcosis 1.

Kulho et al., in 1968 2, among the first to perform polysomnographic recordings in patients with Pickwick syndrome, proposed that the “shunt” of Upper Airways be carried out by means of permanent tracheostomy, in order to exclude the anatomic district in which the obstructive events occurred. This is a surgical proposal which appears extremely “radical” and certainly not completely satisfying for the patient, but that was a pioneering age, in which subjects with morbid obesity and severe grade of OSAS (RDI > 60, SaO2 < 60%) were studied and there were no other instrumental or surgical alternatives (CPAP).

The surgical procedure proposed by Kulho consisted in a classic tracheostomy with the positioning of a fenestrated cannula, maintained closed during the day to allow phonation 2. Albeit, the merits of permanent tracheostomy should be recognised: it has unequivocally demonstrated the obstructive cause of clinical patterns previously interpreted differently, it was the first surgical procedure proposed to solve these apnoeas and is still the only surgical option which ensures, even in very severe cases, complete disappearance of apnoeas and thus remission of the clinical pattern.

When a permanent tracheostomy is necessary, the most interesting surgical technique is the so-called Skin-lined procedure, described by Mayer and Pentta in the Sixties and proposed in conditions of chronic severe respiratory failure with indications for tracheostomy (myasthenia gravis, lateral amyotrophic sclerosis (LAS), severe emphysema, etc.) 3,4. In 1977, Fee and Ward were the first to use Skin-lined Tracheostomy (SLT) in patients with OSAS in order to reduce the risk of development of granulation tissue at tracheal stoma level and improve management of the cannula by the patient him/herself 5.

The efficacy of permanent tracheostomy is demonstrated by reports in the literature. Motta et al., comparing polysomnographic parameters in 6 patients (cardiac frequency, systemic and pulmonary blood pressure, SaO2), before and after tracheostomy, observed a significant improvement in all haemodynamic parameters 6. Partinen et al. studied a population of 198 patients with severe OSAS, for 8 years, of whom 71 submitted to tracheostomy and 127 only to treatment for weight loss. After 5 years, the death rate was 11% in the 127 patients who had opted for conservative therapy vs. no deaths in the patients submitted to tracheostomy 7. Haapaniemi et al., in a longitudinal study on 7 obese patients with severe OSAS submitted to tracheostomy, observed an improvement in the clinical pattern as well as in post-operative polysomnographic findings which remained stable after 5 years 8. Despite the current availability of numerous and heterogeneous therapeutic options, both surgical and non-surgical, for the treatment of OSAS, permanent tracheostomy still has specific indications even if limited to selected cases of severe obstructive apnoeas.

Indications

Permanent tracheostomy (tracheostomy) is used in extreme conditions, as far as concerns the range of options for snoring surgery, and is indicated only in rare conditions of life-threatening OSAS and in which nCPAP is ineffective or not tolerated 9, such as, for example:

- severe hypoxemia (<60%);
- severe hypercapnia;
- severe RDI (>50);
- severe OSAS-correlated arrhythmias (bradycardia, asystolia, ventricular tachycardia, etc.);
- severe Excessive Daytime Sleepiness (EDS);
- ischaemic cardiopathy exacerbated by OSAS;
- ischaemic encephalopathy exacerbated by OSAS;
- obstructive pneumopathy exacerbated by OSAS;
- severe OSAS with little possibility of solution with other surgical procedures, or failure of the latter.

Of the indications, apart from OSAS (besides severe laryngeal or tracheal stenosis, laryngeal diplegias, myasthenia gravis, SLA, intractable aspiration, severe emphysema), in our experience, multisystemic atrophy has become increasingly indicated, due to severe nocturnal laryngospasm with risk of sudden death during sleep.

Therapeutic tracheostomy has to be permanent (tracheostomy) and, therefore, preferably using a specific technique (SLT), able to guarantee:

- greater stability (the presence of an epithelialised bridge between the anterior cervical skin and the tracheal mucosa prevents retraction scarring which leads to spontaneous closure without a cannula, as occurs for all non-skin-lined tracheostomies);
- less risk of granulation tissue (the seal provided
by the skin-mucosa continuity is the best guaran-
tee of a slight, but not absent, tendency of the
peristomal connective tissue to penetrate into the
lumen in the form of granulation tissue, with all
the practical consequences, as well as difficulties
in management, of the case);

• **larger opening of tracheostomy:**
  – for easier and safer management of the cannula in the home setting (removal, cleaning, reinsertion);
  – to allow the stoma to be maintained without a cannula, particularly in the daytime hours, without the risk of untoward stenosis during the day;
  – to allow safe use of personalised obturator, “custom”, to be used, instead of the cannula and, exclusively during the hours the patient is awake, for better phonation (an improved solution compared to simple closure of the cannula which often gives rise to a consistent output of peristomal air);

• **sufficient reversibility**, if necessary, due to perfect preservation of the parietal structure of the trachea.

The only real limit of this elegant technique with cutaneous flaps is that, due to the complex procedure, it would not appear feasible under local anaesthesia, an aspect which cannot be overlooked since these patients often present problems of difficult intubation and/or high risk related to anaesthesia.

**Surgical technique**

SLT, according to the classic technique described by Fee and Ward \(^5\) (with minimal modifications), was carried out in all cases with indications for permanent tracheostomy for therapeutic purposes. General anaesthesia was used in every procedure on account of the complex technique, the long and delicate manipulation of the tracheal structures and the not infrequent severe obesity with a consequently more difficult approach to the tracheal wall for which a similar procedure is not feasible under local anaesthesia.

• **Positioning** of the patient is the same as that of standard tracheotomy, with a pillow under the shoulders and neck which is hyperextended and not turned. To reduce the venous congestion of the neck, it is advisable to position the operating table with the trunk raised by at least 20°. Only in one case was it necessary to tape the chin of the patient to the table in order to guarantee hyperextension of the head in a particularly obese patient. Furthermore, it was necessary, due to severe obesity, to use this same method to anchor extremely voluminous breasts which would otherwise have obstructed the operating field.

• A dermographic pencil is then used to mark the **incisions** in the anterior cervical region. The first step to be decided is the area from which the skin flaps should be harvested, bearing in mind the distance between the jugular cavity and the cricoid cartilage. The positioning of the flaps should take into consideration the following needs, at times contrasting:
  – convenience for the patient to have a stoma as low and less visible as possible;
  – need for the surgeon to work in the subisthmic site;
  – need for the surgeon to work on a trachea which is not yet fully positioned in the mediastinum;
  – need to accommodate the drawing of the flaps in a sometimes minimal cricoid-jugular space;
  – care to bear in mind that when the neck returns from the extended, to the normal position, the incision line tends to slip below.

The final choice is a compromise since the surgeon will have to reflect upon all these aspects also bearing in mind personal experience.

Drawing of the incisions will include (Fig. 1):
  – two parallel transverse lines which are, uppermost, slightly concave, 2 or 3 cm in length, 1-1.5 cm apart;
  – a median vertical line which joins the two transverse lines, and thus 1-1.5 cm in length.

The design is virtually a capital “H” rotated 90° with respect to the vertical position.

The length of the incisions is empirically calculated in proportion to the adipose neck tissue. In the case of a particularly voluminous neck, a longer flap is necessary to reach, through thicker adipose tissue, the trachea which is relatively further away from the skin layer.
Infiltration of the incision lines is then carried out with anaesthetic and vasoconstrictor. The operative field is disinfected and delimited. A n. 15 cryo-bistoury is used for the incisions, in order to lift, from the superficial cervical bundle, two skin flaps together with the subcutaneous tissue. The flaps are then positioned laterally with two sutures on each side (Fig. 2). Fatty tissue is then removed from the flaps, taking care to remove all the adipose tissue from the distal tips, progressively leaving a larger amount of fat as the root of the flap itself is approached, this should ensure the maximum afferent and efferent vascularisation possible.

Dissection along the alba linea as far as the peritracheal fascia is then carried out. Management of the thyroid isthmus is, in effect, related to the opening in the anterior tracheal wall. If sufficient, the isthmus is dislocated upwards. More frequently, it is necessary, rather than with a median section, to perform a true isthmectomy, which allows ample exposure of the tracheal axis. At this stage, use of strong, flat and sufficiently long dilators offers optimal exposure placing, uppermost, 2 on each side (Fig. 3) and, possibly, a third in correspondence to the jugular cavity.

Incision of the two tracheal flaps, superior and inferior, is then performed. To this end, in the centre of the open tracheal area, three “H” incisions are made, respectively, two paramedian vertical parallel incisions, 2.5 cm in height, symmetrical, 1.5 cm one from the other. Incisions are joined 1 cm from the upper border by a third horizontal tracheal section, 1.5 cm in length. The final result is of two upper and lower adjacent tracheal flaps; the lower flap, which is deeper with respect to the skin, is 0.5 cm longer than the upper flap. These two flaps are also sutured at the corners and pulled to the outside (Fig. 4).

Haemostasis is then checked and final suturing of the flaps is performed. In this procedure, the two skin flaps, right and left, meet the two upper and lower tracheal flaps, sealing the opening with a continuous skin bundle. This is achieved by suturing first the upper and lower tracheal flaps with the respective skin (Fig. 5). Each border of the skin flap is then carefully sutured to the respective lateral walls of the tracheotomic opening (Fig. 6). Two more stitches are introduced to join the borders of the flaps together, completely isolating the subcutaneous and connective tissue of the neck from the surface (Fig. 7). At this stage, the tracheal tube is removed and the trachea cannula is introduced.

A cuffed tracheal cannula is then introduced (in the present case, a Shiley n. 8) for the first 3-4
days, after which an uncuffed fenestrated cannula of the same diameter is used (unlike in standard tracheotomies, the well-known effect favouring granulation due to fenestration is improbable due to epithelialisation of the stoma).

• Within approximately 1 month, stabilisation of the tracheotomy is achieved, which is concentrically narrowed (about 1/3 of the original diameter). At this stage, if the local stomal condition and the social-educational level of the patient and his family allow, it is possible:
  – at night, to leave the tracheostoma uncovered and functioning, with or without the cannula.
  – during the day:
    – to cover the stoma with a waterproof plaster;
    – or to close the stoma with a cannula and its button;
    – or close the stoma with a silicone “custom” obturator or dental plaster cast paste.

Materials and methods

A retrospective study has been carried out on personal experience with the use of SLT.

A total of 1103 ronchosurgical procedures were carried out at the ENT Division of Forlì, between September 1996 and April 2002, on the different areas of the upper aero-digestive pathways in 530 patients, mean age 50 years (range 26 - 78), with a male prevalence (81%), presenting variable degrees of OSAS severity:

• 48 (9%) Simple Snoring (SS).
• 58 (11%) Upper Airway Resistance Syndrome (UARS).
• 424 (80%) Obstructive Sleep Apnoea Syndrome (OSAS), of which:
  – 79 (15%) mild (RDI 10-20);
  – 170 (32%) moderate (RDI 21-40);
  – 175 (33%) severe (RDI 41-60) or very severe (RDI > 60).

A total of 472 patients (89%) were operated upon under general anaesthesia, while 58 (11%) received local anaesthesia. Furthermore, an evaluation was made of those cases submitted to SLT for the treatment of non-OSAS sleep-related respiratory disorders.

Results

A total of 10 SLTs were performed.

Indications were as follows:

• 7 patients presented nocturnal laryngeal stridor, a form of laryngospasm with severe respiratory insufficiency due to a very rare neurological disease, multisystemic atrophy or MSA. These cases
3 patients had a polysomnographic diagnosis of severe OSAS (mean RDI 67.3, mean BMI 37.3) and intolerance to nCPAP:
- in the first case (D.A., a 42-year-old female, BMI 31, RDI 68, with congenital cerebroathy, SLT was carried out following failure of the multisite procedure (turbinoplasty, UPPP and antero-inferior hyoid suspension) according to Stanford phase 1 and refusal of maxillo-mandibular advancement;
- the second OSAS patient, submitted to SLT (L.A., a 60-year-old male), appeared to be severely symptomatic (RDI 75), severely overweight (BMI 44), with exaggerated bradytype (height 150 cm, cervical circumference 48 cm) and presented severe hypothyroidism, not previously detected. The extremely unfavourable anatomical conformation suggested great caution in advising multi-site surgical options, the need for medical treatment (diet, thyroid hormone) resulted, however, in performing surgery in stages, while the severe conditions required immediate treatment: SLT was, therefore, carried out. After 18 months, thyroid replacement therapy with a weight loss of 17 kg (BMI 37), a Tongue-Base Reduction with Hyoepiglottoplasty (TBRHE) according to Chabolle was carried out, the next month UPPP with tonsillectomy was performed and, after only one month, closure of the SLT was effected;
- the third OSAS patient (M.A., a 50-year-old male, BMI 37, RDI 59), submitted to SLT, was, therefore, carried out. After 18 months, thyroid replacement therapy with a weight loss of 17 kg (BMI 37), a Tongue-Base Reduction with Hyoepiglottoplasty (TBRHE) according to Chabolle was carried out, the next month UPPP with tonsillectomy was performed and, after only one month, closure of the SLT was effected;

Outcome was favourable in all 10 cases:
- elimination of apnoea in patients with neurological indications (the 3 deaths occurring in the 7 patients with multisystemic atrophy were due to fatal progression of the disease);
- almost complete disappearance of apnoea (RDI <10) and related symptoms (immediate and astonishing effects upon sleep and daily symptoms) in the OSAS patients (patients with massive obesity and a variable degree of hypoventilation of the restrictive type);

The tracheal cannula was almost always necessary to guarantee adequate opening of the tracheostomy in any sleeping position, particularly in those patients presenting OSAS due to a cervical conformation (short and wide circumference) and the large skin plications due to abnormal fat tissue. Management of the cannula, this was replaced by a Shiley cannula in the home setting did not present any difficulty, as in total laryngectomy. For better phonation, and only whilst awake, it is advisable to remove the cannula and to apply, with the use of adhesive plaster, the personalised obturator, which is obtained by shaping the plaster used for dental casts on the tracheostomy tract, thus obtaining a cone shaped obturator. This solution was found to be better than simple closure of the cannula (which often gives rise to consistent output of peristomal air), which is fairly well accepted and used in various ways.

As for any tracheotomy, SLT was poorly tolerated, differing according to the pathological condition. Thus, while the severity and possible evolution of multisystemic atrophy did not give rise to any particular problem to adapt to SLT, the situation, in our experience, was quite different in OSAS patients:
- due to difficulties encountered by the family of the congenital cerebropathy patient, in the management of the cannula, this was replaced by a Montgomery T tube. A few days after the latter discharge, the patient was admitted to the Intensive Care Unit due to an acute nocturnal hypoxic coma and saved by the physician on duty thanks to timely reintroduction of a Shiley cannula in the periventricular coma. With the use of mechanical ventilation, the patient regained the conditions prior to this episode, without any apparent adverse effects, within 2 days. Following this experience, the patient and the family take greater care in the management of the stoma and cannula;
- the second patient (L.A.), whilst pleased with the
improvement in the post-SLT psycho-physical conditions, very much wanted (and awaited!) a medical and surgical solution that would probably lead to closure of the tracheotomy (which, indeed, occurred after 18 months);

• the third patient (M.A.), three months after SLT, personally removed the cannula at home, but had to return to our Centre due to partial stenosis of the tracheostomy; a few days after progressive dilatation, by means of insertion of a cannula with gradually increasing diameter, the patient suddenly removed the cannula again and left the hospital against the judgement of the physicians (during the days prior to this episode, the patient had expressed the intention to undergo abdominal surgery for obesity, a solution which, moreover, had met with the approval of our group).

Conclusions
Better understanding of OSAS and the increase in treatment options available (instrumental, medical and surgical) it is, nowadays, extremely rare to encounter cases in which tracheotomy is the only feasible treatment. These conditions occur only in severe OSAS, at high risk (even life-threatening) and in which n-CPAP is not tolerated or ineffective (in our experience, only 1.7% of severe, or very severe, OSAS submitted to surgical treatment).

In these selected cases, tracheotomy is carried out using the skin-lined technique, which offers several advantages, such as a greater opening of the stoma, higher stability over time, less risk of granulation tissue, reversibility. The procedure did not appear to give rise to important complications. The expected functional results were achieved with immediate disappearance of daytime symptoms and satisfactory recovery as far as concerns nocturnal apnoeas. This technique would be suitable for use in patients with sleep disordered breathing of neurological origin (laryngeal stridor due to multisystemic atrophy). Besides sleep-related disorders, various clinical conditions for which permanent tracheotomy is indicated, may benefit from the skin-lined technique, such as, for instance, severe laryngeal or tracheal stenoses, laryngeal diplegia, miasthenia gravis, LAS, intractable aspiration and severe emphysema.

References

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