Medical Interventions for
Spasmodic Dysphonia & Some Related Conditions:
A Systematic Review

Technical Report 2

Academy of Neurologic Communication Disorders and Sciences

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## Medical Interventions for Spasmodic Dysphonia & Some Related Conditions

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Medical Interventions for Spasmodic Dysphonia & Some Related Conditions

BACKGROUND

This systematic review summarizes the results of one segment of the Practice Guidelines for Dysarthria that are being developed through the ANCDS and co-sponsored and funded by ASHA, through the office of the VP of Clinical Practices in Speech-Language Pathology, and from the Steering Committee of Division 2. Additionally, co-sponsorship and funding support were received from the Department of Veterans Affairs (DVA). The review addresses surgical, pharmacologic, and medical approaches to managing voice/laryngeal abnormalities associated with dysarthrias due to movement disorders (e.g., dystonia, tremor), disorders that fall under the broad heading of hyperkinetic dysarthrias. Behavioral management for respiratory/phonatory dysfunction in dysarthria is not addressed here, but this topic will be reviewed in a future module. Other topics related to laryngeal dysfunction, such as medical treatments for neurologic voice disorders secondary to laryngeal weakness or paralysis also will be reviewed in future modules.

The primary focus of this effort was to review the literature on the management of spasmodic dysphonia (SD); some information about management of voice tremor and movement disorders affecting non-laryngeal speech structures (e.g., lingual dystonia) was also compiled. Much of the evidence reviewed focused on recurrent laryngeal nerve (RLN) section and the use of botulinum toxin (BT) injections for the management of SD. This documentation was reviewed by members of writing committee along with a panel of experts. These experts (N = 30) were volunteer members of ANCDS or Speech-Language Pathologists who had published in the area of management of SD. Most of the panel of experts (73%) hold doctoral degrees and has substantial clinical experience (mean duration of practice: 19.4 years).

RATIONALE FOR THE REVIEW

The following section will discuss factors that motivated this review. Because most of the evidence summarized here focuses on SD, a brief explanation of the disorder may help to establish its relationship to the dysarthrias and its relevance to practice guidelines.

The cause of SD is unknown. Just a few decades ago most clinicians believed it was the result of psychological disturbances, such as conversion disorder or musculoskeletal tension driven by psychological stress. However, most clinicians and researchers today have concluded that the disorder most often reflects an underlying involuntary neurologic movement disorder such as dystonia or essential voice tremor. Although some clinicians continue to recognize a psychogenic form of SD (Aronson, 1990), most place such psychogenic voice problems into categories separate from SD (e.g., conversion dysphonia/aphonia, musculoskeletal tension dysphonia). It is not the intent of this report to address these nosologic issues, but the reader should know that the treatments reviewed here have been directed, in the great majority of studies, at voice and speech disorders assumed to be
neurological as opposed to psychological in origin. It is also important to recognize that the existence of voice disorders with clinical perceptual features that resemble those of neurologic SD makes the distinction between neurologic SD and non-neurologic voice disorders essential to management decision making.

Although uncommon, SD is estimated to affect 30,000-50,000 people in North America. More women than men are affected. Onset is most often in the 5th – 6th decade. The adductor variety of the disorder (in which voice is typically continuously or intermittently strained, sometimes to the point of voice stoppages) is far more common (about 90% of cases) than the abductor variety (in which the voice is typically intermittently breathy or aphonie). Some individuals appear to have a combination of adductor and abductor forms. The disorder often results in occupational, social, and emotional problems, with disruption of socioeconomic success and quality of life. Although SD may be accompanied by evidence of dystonia or tremor elsewhere in the body (e.g., torticollis, focal cranial dystonias), in a substantial number of people the abnormal movements are largely confined to the larynx, and are apparent only during speech (i.e., a focal, action-induced movement disorder). Thus, unlike management for many types of dysarthria, efforts to manage SD focus predominantly on only one speech subsystem. Although this review does not address behavioral management, the literature on medical management has been driven at least partly by a conclusion that behavioral intervention, alone, for SD (at least its neurologic forms) is ineffective or suboptimal.

This review was considered important because the frequent misdiagnosis of SD, its interdisciplinary demands for diagnosis and management, its apparent historic resistance to behavioral voice therapy, and the relatively recent development of accepted medical interventions for it make it particularly important for speech-language pathologists to have up-to-date knowledge about the efficacy and effectiveness of medical interventions for the disorder. This information should assist speech-language pathologists in their counseling and management decision making for patients with SD and related hyperkinetic speech disturbances.

This writing committee recognizes that this report might not be comparable in its method of evaluation or criteria for drawing conclusions about treatment effects to those that might be developed by our colleagues in neurology or otorhinolaryngology. We suspect, however, that any differences that might emerge among disciplines will not be great, and hope that any differences will promote further study that benefits the individuals we all strive to serve.

THE LITERATURE SEARCH

Because this review focused on medical interventions, the MEDLINE database - covering articles from 1966 through December 2000 - was the primary search vehicle. The initial search used the key words “spasmodic dysphonia” and “spastic dysphonia” to identify appropriate citations. The search was limited to human studies reported in English. The search yielded approximately 250 citations. The titles were then reviewed and those that suggested any focus on management were identified. This yielded 133 citations whose
abstracts (when available) were reviewed. Letters to the editor, articles that only non-
critically summarized other work, articles whose abstracts clearly indicated they did not
address management, and articles focused on management of problems that clearly were not
neurologic in origin were then excluded.

An additional MEDLINE search using key words “essential voice tremor,” “voice tremor,”
and “organic voice tremor” yielded an additional four relevant articles that were not redundant
with those identified during the search on SD.

The PsycINFO database was then reviewed in a similar manner. Only one additional relevant
article was identified. A small number of additional articles were identified through reviews
of texts and other publications, and from comments from expert reviewers.

Eight relevant or possibly relevant articles could not be obtained for review. Their abstracts,
titles, or publication dates suggest that their inclusion would not substantively alter the
conclusions derived from the reviewed evidence. These citations are provided in the reference
section so readers can judge the likely impact of their omission.

The available articles were divided into three categories. Each article (never just the abstract)
was then reviewed by two individuals (JD & KY) using guidelines for evaluating evidence
established by the Writing Committee (Yorkston et al., in press-a, in press-b). The categories
include:

1) SD - RLN section = 24 references
2) SD - BT treatment = 63 references
3) Miscellaneous Medical Treatments = 25 references

Three Evidence Tables are summarized in subsections that follow. The first two – RLN
section and BT treatments for SD – are each reviewed in some detail because the amount of
evidence regarding them is substantial, and because they are fairly well defined treatments.
The third table of evidence – miscellaneous medical treatments for SD and related conditions
– is quite heterogeneous in the treatments covered and not as amenable to concise summary.

RECURRENT LARYNGEAL NERVE (RLN) SECTION FOR SD

Recurrent Laryngeal Nerve (RLN) section was first reported as a treatment for adductor SD in
1976. Following this procedure, the paralyzed vocal fold assumes a paramedian position so
that the remaining vocal fold cannot closely approximate it.

Intervention Studies

A number of articles describing the results of RLN section have appeared, with most
published in the 1970s and 1980s (Table 1 and Evidence Table 1). This publication
chronology generally coincides with the initial popularity of RLN section, and then its decreased use as consensus emerged that the more recently developed BT injection was the preferred medical treatment for SD in many cases.
Table 1. Publication dates of RLN treatment articles.

<table>
<thead>
<tr>
<th>Time Frame</th>
<th># of Intervention Articles</th>
</tr>
</thead>
<tbody>
<tr>
<td>&lt; 1976</td>
<td>0</td>
</tr>
<tr>
<td>1976 – 1979</td>
<td>5</td>
</tr>
<tr>
<td>1980 – 1989</td>
<td>17</td>
</tr>
<tr>
<td>1990-1999</td>
<td>2</td>
</tr>
<tr>
<td>2000</td>
<td>0</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>24</strong></td>
</tr>
</tbody>
</table>

Subjects

The total number of subjects with SD reported in the 24 identified articles is 1,324. This figure is artificially inflated misleading because several authors/institutions have published two or more studies containing progressively cumulative data. If only the largest N study by authors from given institutions is counted, the lowest estimate of the total number of different subjects studied is about 395. Nonetheless, this number is substantial in comparison to treatment studies of dysarthria that have used behavioral interventions.

The percentage of studies reporting specific subject descriptors is summarized in Table 2. It should be noted that not all subject characteristics are equally important or necessarily relevant to SD or its treatment with RLN section. For example, diadochokinesis, sensation, SES/education, and hearing/vision would not generally be considered essential information.

Table 2. Percentage of studies reporting specific subject descriptors.

<table>
<thead>
<tr>
<th>Subject Characteristics</th>
<th>% Of Studies Reporting</th>
</tr>
</thead>
<tbody>
<tr>
<td>Medical Diagnosis</td>
<td>88</td>
</tr>
<tr>
<td>Type of dysarthria</td>
<td>88</td>
</tr>
<tr>
<td>Age</td>
<td>68</td>
</tr>
<tr>
<td>Gender</td>
<td>68</td>
</tr>
<tr>
<td>Speech characteristics</td>
<td>60</td>
</tr>
<tr>
<td>Treatment history</td>
<td>52</td>
</tr>
<tr>
<td>Severity of dysarthria</td>
<td>52</td>
</tr>
<tr>
<td>Disease severity</td>
<td>52</td>
</tr>
<tr>
<td>Time post onset</td>
<td>48</td>
</tr>
<tr>
<td>Medications</td>
<td>32</td>
</tr>
<tr>
<td>Acoustic data</td>
<td>24</td>
</tr>
<tr>
<td>Neurologic examination data</td>
<td>20</td>
</tr>
</tbody>
</table>
Physiologic data | 16
SES or education | 8
Cognition/language | 0
Hearing or vision | 0
Sensation | 0
Diadokokinesis | 0

Types of Studies

The type of study varied (Table 3), and a number of group studies also included case descriptions. Eleven articles represented group studies, only 2 of which included a comparison group (normal controls). Seventeen studies represented case/case series; five of them were presented as part of a group study. Only one study used a single subject design.

Table 3. Type of study. Numbers and percentages sum to > 24 and >100%, respectively, because some studies fell into more than one category.

<table>
<thead>
<tr>
<th>Type of Study</th>
<th>Articles (Percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Case/Case Series</td>
<td>17 (71%)</td>
</tr>
<tr>
<td>Single Subject</td>
<td>1 (4%)</td>
</tr>
<tr>
<td>Group (total)</td>
<td>11 (44%)</td>
</tr>
<tr>
<td>Group (comparison study)</td>
<td>2 (8%)</td>
</tr>
<tr>
<td>Total # of relevant articles</td>
<td>24</td>
</tr>
</tbody>
</table>

The treatment described was considered replicable in 68% of the studies (Table 4). It should be noted that this percentage reflects the adequacy of the description of the RLN section procedure. In a substantial percentage of studies, the method for measuring the effect of the treatment was not considered replicable. The reasons for this are reflected in ratings of psychometric adequacy, evidence of control, or the rater’s comments in the Table of Evidence.

Table 4. Replicability of treatment (a few studies fell into more than one category).

<table>
<thead>
<tr>
<th>Replicability</th>
<th>Articles (Percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Replicable</td>
<td>17 (68%)</td>
</tr>
<tr>
<td>General information</td>
<td>6 (24%)</td>
</tr>
<tr>
<td>Incomplete</td>
<td>1 (4%)</td>
</tr>
</tbody>
</table>

Outcome Measures

Across the studies reviewed, outcome measures focused on impairment, activity limitation, and participation restriction. A substantial majority of studies focused on measures that were
used to determine impairment. Measures of impairment most often included: perceptual ratings and general clinical descriptions; patient and clinician ratings; acoustic analyses; visualization of the larynx; electroglottography, electromyography; and aerodynamic measures. Measures of activity limitation were less frequently employed than measures of impairment. They most often included patient self-ratings, clinician ratings, and general descriptions; ratings most often focused on general improvement or reduced severity, ability to communicate, ease of communication, reduction of preoperative limitations, and changes in positive and negative traits. Measures of participation restriction were infrequently reported (~9% of the studies). They tended to rely on general descriptions and patient testimonial comments.

There was considerable variability across studies relative to psychometric adequacy and evidence for control. Many studies were adequate in one of those characteristics, but only a minority was adequate in both. Many of the studies did explicitly address risks and complications.

Levels and Quality of Evidence

Data-based treatment studies were categorized according to levels and quality of evidence. Quality of evidence ratings used criteria employed by the American Academy of Neurology Quality Standards Subcommittee for developing practice parameters (1999) in which studies are rated as Class I (prospective, randomized, controlled clinical trial with masked outcome assessment, in a representative population), Class II (prospective, matched group cohort study in a representative population with masked outcome assessment that meets Class I criteria, or a randomized controlled trial that lack one criteria for Class I), Class III (all other controlled trials, including well-defined natural history controls, in a representative population, where outcome assessment is independent of patient treatment), or Class IV (evidence from uncontrolled studies, case series, case reports, or expert opinion). Studies were also categorized for Levels of Evidence (Beukelman, 2001), in which they were rated as Authoritative (expert opinion from professionals, consumers, secondary consumers), Experimental (randomized controlled studies, studies with controls, single subject studies), or Observational (case studies, series of case studies, qualitative studies, structured behavioral observations).

Table 5 summarizes these ratings for RLN studies. There have been no Class I or II studies of this treatment. AAN guidelines indicate that one convincing Class I study and two consistent Class II studies justify a conclusion that a treatment is “established as effective.” At least one convincing Class II study or at least three consistent Class III studies justify a conclusion that a treatment is “probably effective.” At least two convincing and consistent Class III studies are required for a treatment to be considered “possibly effective.” Class IV studies, even if consistent, require a conclusion that a treatment is unproven. According to these criteria, the data suggest that RLN section for SD should be considered possibly effective or unproven, especially when inconsistencies within and among studies are considered (inconsistencies
represented in long term effectiveness data). The levels of evidence data (Beukelman criteria) (Beukelman, 2001) indicate that most (86%) of the studies were observational.

Table 5. Summary of levels and quality of evidence for studies of RLN section for SD. (22/24 articles were data-based).

<table>
<thead>
<tr>
<th>AAN Criteria</th>
<th># of Studies</th>
<th>Beukelman Levels</th>
<th># of Studies</th>
</tr>
</thead>
<tbody>
<tr>
<td>Class I</td>
<td>0</td>
<td>Experimental</td>
<td>3</td>
</tr>
<tr>
<td>Class II</td>
<td>0</td>
<td>Observational</td>
<td>18</td>
</tr>
<tr>
<td>Class III</td>
<td>4</td>
<td>Authorative</td>
<td>1</td>
</tr>
<tr>
<td>Class IV</td>
<td>18</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Conclusions: RLN Section

Treatment Outcomes

The following conclusions about RLN Section as a treatment for SD appear to be justified by the weight of the evidence reviewed:

- RLN Section as a treatment for adductor SD results in a substantial degree of improvement for a substantial percentage of patients, but in spite of permanent vocal cord paralysis in most cases, its long-term effectiveness appears to be limited. In general, the short-term effectiveness of RLN section is reflected across measures showing less impairment, less activity limitation, and less participation restriction, and the improvement resulting from the treatment is generally evident to patients as well as their treating physicians and speech-language pathologists. However, recurrence of SD signs and symptoms is common (ranging across studies from a minority to a substantial majority of cases within 3 years), with higher recurrence rates in women than in men. Laser thinning of the paralyzed vocal cord, as a secondary management procedure, when recurrence occurs, appears effective for some patients. It also should be noted that refinements or variations of nerve section procedures for SD have been developed in recent years, and that they may prove to be more effective than traditional RLN section. These treatments are discussed in the section of this report that addresses miscellaneous medical treatments (recurrent laryngeal nerve avulsion, selective laryngeal adductor denervation-reinnervation).

- RLN Section (or refinements of it, such as RLN avulsion or selective laryngeal adductor denervation-reinnervation) for adductor SD may be appropriate for some individuals who
fail to respond or receive less than desired benefits from BT injection, or who prefer to pursue the possibility of a longer lasting or “permanent” treatment for their adductor SD.

Side Effects

Regarding side effects, RLN is intended to produce vocal cord paralysis, and that paralysis is usually permanent. Common adverse effects include weak-breathy-hoarse voice quality and dysphagia, both of which are usually temporary. Respiratory distress occurs occasionally. Other adverse effects are uncommon. As already noted, recurrence of symptoms of SD is frequent.

BOTULINUM TOXIN (BT) INJECTION FOR SPASMODIC DYSPHONIA

Botulinum toxin (BT) is a protease that blocks the release of acetylcholine from the nerve terminals. BT gained full FDA approval for treatment of strabismus, blepharospasm, and VIIth cranial nerve disorders in 1980. First reports of “off-label” use in individuals with SD appeared in the later 1980s. For an excellent review of clinical uses of BT, see Blitzer & Sulica (2001).

Intervention Studies

Numerous articles describing BT treatment for SD have appeared during the past 15 years. BT is a relatively new treatment for SD and the vast majority of articles have been published during the past decade (Table 6 and Evidence Table 2).

Table 6. Publication dates of BT treatment articles.

<table>
<thead>
<tr>
<th>Time Frame</th>
<th># of Intervention Articles</th>
</tr>
</thead>
<tbody>
<tr>
<td>&lt; 1980</td>
<td>0</td>
</tr>
<tr>
<td>1980 – 1989</td>
<td>5</td>
</tr>
<tr>
<td>1990 – 1999</td>
<td>50</td>
</tr>
<tr>
<td>2000</td>
<td>8</td>
</tr>
<tr>
<td>Total</td>
<td>63</td>
</tr>
</tbody>
</table>

Subjects

The total number of subjects with SD reported in the 63 identified articles is 4,272. This figure is artificially inflated, however, because several authors/institutions have published two or more studies containing progressively cumulative data. If only the largest N of the studies by authors from given institutions is counted, the lowest estimate of the total number of different subjects studied is probably between 1,425 and 1,525. Nonetheless, this number is substantial, especially in comparison to treatment studies of dysarthria that have used behavioral interventions.
The percentage of studies reporting specific subject descriptors is summarized in Table 7. It should be noted that not all subject characteristics are equally important or necessarily relevant to SD or its treatment with BT injection. For example, diadochokinesis, sensation, SES/education, and hearing/vision would not generally be considered essential information.
Table 7. Percentage of studies reporting specific subject descriptors.

<table>
<thead>
<tr>
<th>Subject Characteristics</th>
<th>% Of Studies Reporting</th>
</tr>
</thead>
<tbody>
<tr>
<td>Type of dysarthria</td>
<td>100</td>
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<tr>
<td>Medical Diagnosis</td>
<td>97</td>
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<tr>
<td>Medications</td>
<td>86</td>
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<tr>
<td>Age</td>
<td>89</td>
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<tr>
<td>Gender</td>
<td>89</td>
</tr>
<tr>
<td>Speech characteristics</td>
<td>71</td>
</tr>
<tr>
<td>Treatment history</td>
<td>62</td>
</tr>
<tr>
<td>Time post onset</td>
<td>59</td>
</tr>
<tr>
<td>Severity of dysarthria</td>
<td>52</td>
</tr>
<tr>
<td>Disease severity</td>
<td>48</td>
</tr>
<tr>
<td>Acoustic data</td>
<td>41</td>
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<tr>
<td>Physiologic data</td>
<td>40</td>
</tr>
<tr>
<td>Neurologic examination data</td>
<td>37</td>
</tr>
<tr>
<td>SES or education</td>
<td>3</td>
</tr>
<tr>
<td>Sensation</td>
<td>2</td>
</tr>
<tr>
<td>Cognition/language</td>
<td>0</td>
</tr>
<tr>
<td>Hearing or vision</td>
<td>3</td>
</tr>
<tr>
<td>Diadokokinesis</td>
<td>0</td>
</tr>
</tbody>
</table>

Types of Studies

The type of study varied (Table 8), and a number of group studies also included case descriptions. Fifty-three articles represented group studies, 19 of which included a comparison group. Group comparison studies included one with a double-blind, randomized, placebo control design. Others varied considerably in the nature of comparisons made; for example, comparison of SD speakers to normal speakers, comparison of unilateral to bilateral injections, comparison of percutaneous to transoral injections, comparison of results as a function of dose and site of injection, and comparison of effects of BT to BT plus voice therapy. Most of the studies were retrospective studies of clinic populations. Twenty-one studies represented case/control series; 13 of them were presented as part of a group study. Only three studies used a single subject design.

Table 8. Type of study. Numbers and percentages sum to > 63 and > 100%, respectively, because some studies fell into more than one category.
The treatment described was considered replicable in 83% of the studies (Table 9). It should be noted, however, that this impressive percentage primarily focused on the description of the BT injection procedure. In a substantial number of studies, the method for measuring the effect of the treatment was not considered replicable. The reasons for this are reflected in ratings of psychometric adequacy, evidence of control, or the rater’s comments in the Table of Evidence.

Table 9. Replicability of treatment. Percentages sum to > 100% because some studies fell into more than one category.

<table>
<thead>
<tr>
<th>Replicable</th>
<th>52 (83%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>General information</td>
<td>16 (25%)</td>
</tr>
<tr>
<td>Incomplete</td>
<td>5 (8%)</td>
</tr>
</tbody>
</table>

**Outcome Measures**

Across the studies reviewed, outcome measures focused on impairment, activity limitation, and participation restriction. A substantial majority of studies focused on measures that were used to determine impairment. These measures of impairment most often included: acoustic analyses; patient and clinician ratings; visualization of the larynx (including videolaryngoscopy and videostroboscopy); electroglottography, electromyography, and aerodynamic measures; auditory perceptual measures. Measures of activity were less frequently employed than measures of impairment. They most often included patient self-ratings and clinician (speech pathologist or physician) ratings; the ratings most often focused on general improvement, ability to communicate, duration of benefit, presence and duration of side effects, and general satisfaction. Measures of participation were not frequently reported (~25% of the studies). They tended to focus on testimonial comments, responses to handicap scales, or description of patients’ social and work limitations prior to treatment and changes thereafter.

There was considerable variability across studies relative to psychometric adequacy and evidence for control, but a number of studies were adequate in one or both of those characteristics. Many of the studies explicitly addressed risks and complications. Study results and conclusions did not appear to differ as a function of the degree to which they met standards for psychometric adequacy and control.
Levels and Quality of Evidence

Data-based treatment studies were categorized according to levels and quality of evidence. Table 10 summarizes these ratings. The levels of evidence data (Beukelman criteria) indicate that about 45% of the evidence is experimental, with the remaining data being observational. There have been one Class I study and four Class II studies. AAN guidelines indicate that one convincing Class I study and two consistent Class II studies justify a conclusion that a treatment is “established as effective.” At least one convincing Class II study or at least three consistent Class III studies justify a conclusion that a treatment is “probably effective.” Each of the Class I and II studies concluded that botox injection was effective, although the number of Ss in each study were relatively small and the specific treatments (in terms of injection site, dose, etc.) varied. It is important to note that every study reviewed reported beneficial effects of the injection for a significant proportion of patients. Thus, these data suggest that BT injection for SD is effective or probably effective according to the AAN criteria.

Table 10. Summary of levels and quality of evidence. Only 58/63 articles were data-based.

<table>
<thead>
<tr>
<th>AAN Criteria</th>
<th># of Studies</th>
<th>Beukelman Levels</th>
<th># of Studies</th>
</tr>
</thead>
<tbody>
<tr>
<td>Class I</td>
<td>1</td>
<td>Experimental</td>
<td>25</td>
</tr>
<tr>
<td>Class II</td>
<td>4</td>
<td>Observational</td>
<td>33</td>
</tr>
<tr>
<td>Class III</td>
<td>17</td>
<td>Authoratative</td>
<td>0</td>
</tr>
<tr>
<td>Class IV</td>
<td>36</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Conclusions: BT Intervention

The following conclusions about BT injection treatment for SD appear to be justified by the weight of the evidence reviewed:

BT Injection for Adductor SD

BT injection as a treatment for adductor SD results in a substantial degree of improvement for a substantial percentage of patients, with benefits generally lasting for three to four months. To date, it appears that injections may be repeated indefinitely in most cases. In general, the positive effects of this treatment are reflected across measures showing less impairment, less activity limitation and less participation restriction. The resulting improvement is generally evident to patients as well as their treating physicians and speech-language pathologists. BT
injection can also be an effective treatment for people who have had RLN section to treat adductor SD, with subsequent return of signs and symptoms of the disorder.

**Side Effects**

There are no known serious side effects of BT treatment for SD. Adverse effects include weak-breathy voice and mild dysphagia that may last for a few days to 2-3 weeks in a substantial percentage of patients. It appears that these side effects can be minimized by adjusting the dose and injection site (e.g., unilateral versus bilateral injection). Other side effects, such as pain at the site of injection or respiratory difficulty, are uncommon.

**BT Injection for Abductor SD**

In comparison to studies of BT injection for adductor SD, studies of BT injection for abductor SD are fewer in number, the number of subjects reported is considerably smaller, and the level and quality of evidence are less adequate. Although the one study that has employed blinded auditory-perceptual and fiberscopic ratings failed to demonstrate significant change post BT treatment (Bielamowicz et al 2001), the remaining studies have consistently reported beneficial outcomes for a majority of patients on the basis of unblinded clinician judgment or patient report (a majority of patients in the Bielamowicz et al., (2001) study also reported benefits). It thus appears reasonable to conclude that BT injection for abductor SD probably provides subjective patient-perceived benefit for a majority of cases but, in comparison to adductor SD, its effectiveness is less pronounced and occurs in a smaller percentage of patients. Further study of the effectiveness of BT injection for abductor SD, and development of alternative treatments, are clearly necessary.

**Treatment Comparisons**

BT injections for SD vary in terms of dose amount, site (e.g., unilateral versus bilateral), and technique (e.g., percutaneous versus transoral). These differences appear to reflect physician preference and various patient needs and responses. To date, it appears that there is no ideal dose amount that is equally effective for all patients, and that dose probably needs to be established on a case-by-case basis. Unilateral versus bilateral injection and transoral versus percutaneous approaches each have advantages and disadvantages, and there does not appear to be strong evidence or consensus that any injection site or method of injection is clearly superior.

**Accumulating Evidence**

The conclusions drawn here receive some support from a report by the American Academy of Neurology Therapeutics and Technology Assessment subcommittee in 1990 that concluded that “local injections of Botox into the vocalis muscle is accepted as a safe and efficacious modality for the treatment of symptoms of adductor spasmodic dysphonia. Botox therapy is a
promising therapy for the treatment of abductor SD.” (American Academy of Neurology Therapeutics and Technology Assessment Subcommittee, 1990) It should be noted that that report was made at a time when only 98 cases receiving BT injection for abductor SD had been reported. The subsequent evidence reviewed in this report appears to strengthen that committee’s conclusion.

MISCELLANEOUS MEDICAL TREATMENTS

The 25 articles reviewed under this heading were quite heterogeneous. Evidence Table 3 summarizes these studies in chronological order. Some represent treatments that seem to have been abandoned because of lack of effectiveness. Some represent alternatives when BT injection is ineffective, unacceptable to patients, or cannot be used. Others represent treatments that can be considered experimental and, perhaps, promising.

SD – Miscellaneous Surgical Treatments

The following is a brief description of several surgeries that have been used for the treatment of SD.

Laryngeal Nerve Crush

Laryngeal nerve crush for SD has been investigated in a small number of studies as an alternative to RLN Section. The procedure results in initial paralysis of a vocal cord, but vocal cord function eventually returns because of RLN motor fiber regeneration. Although initial results were successful, follow up suggests a very high regression rate. The developers of the procedure (Biller, Som, & Lawson, 1983) concluded that laryngeal nerve crush should not be a recommended treatment for SD; at that time, the authors recommended RLN Section as the preferred treatment for SD.

Recurrent Laryngeal Nerve Avulsion

Recurrent laryngeal nerve avulsion (RLNA) is a relatively new procedure that involves removing the distal nerve up to its insertion into the laryngeal muscle thus limiting the neural regrowth. The effect of RLNA on voice for patients with adductor SD has been investigated in two studies. Complete RLNA should prevent regeneration of nerve more effectively than just sectioning the nerve, as in RLN resection. The authors of the most recent report (Weed et al., 1996) suggest that RLNA may be appropriate for those who do not benefit from or tolerate BT injection. Although it is possible this approach will be more lastingly effective than RLN Section, further study is necessary.

Selective Laryngeal Adductor Denervation-Reinnervation
This is a new surgical procedure for adductor SD that involves bilateral denervation of the recurrent laryngeal nerve with reinnervation of the distal stumps with branches of the ansa cervicalis nerve. It has been developed for patients with severe adductor SD seeking an alternative to Botox injection, and as a procedure likely to have a lower SD recurrence rate than RLN Section. Berke et al. (1999), in a group/case series study, have reported promising results. Replication by others and longer-term follow up are needed.

Laryngeal Framework Surgery

Laryngeal framework surgery, a technique that permits adjustment of the position and tension in the vocal folds, has been used in a small number of subjects with adductor or abductor SD. The results of the two published reports (Isshiki et al., 2000; Tucker et al., 1989), on a limited number of subjects, with limited follow up (less than two years), suggests that laryngeal framework surgery has potential for managing SD in some patients. Data are insufficient, however, to permit a conclusion that laryngeal framework surgery should be routinely recommended for managing SD.

SD – Other Medical Treatments

RLN Stimulation

Vagus nerve or RLN stimulation (a variation on the use of vagus nerve stimulators to control epilepsy) has been used for a small number of patients with SD. Although percutaneous and implanted stimulators have produced voice improvement in a small number of patients, the last report of its use was in 1994. It seems reasonable at this time to conclude that vagus or RLN stimulation has not been demonstrated to be an effective treatment for SD.

Pharmacologic Management

There are infrequent reports of attempts to treat SD with medications that may be effective for controlling tremor or dystonia elsewhere in the body. The limited data do not support predictable effectiveness of any single drug for managing SD.

Biofeedback

A single study has investigated the treatment of SD with EMG biofeedback (Henschen and Burton, 1978). The treatment was unsuccessful.

Chiropractic Manipulation

Chiropractic manipulation has been reported as successful in managing SD in a single patient (Wood, 1991). Subsequent studies have not been published. Thus, data are insufficient to support chiropractic manipulation as an effective management for SD.
Other Movement Disorders: Miscellaneous Medical Treatments

The articles reviewed under this heading represent heterogeneous treatments for a variety of different conditions. A few of these articles also include treatments for SD so they are reviewed both in this and other sections. Some of the treatments are relevant to managing movement disorders that affect laryngeal function, whereas others address movement disorders that affect other levels of the speech system.

BT for Dystonias Other Than SD

BT injection has been used to treat jaw, lingual and other orofacial dystonias. The American Academy of Neurology Therapeutics and Technology Assessment Committee in 1990 reported that 82 published studies have used BT injection for oromandibular dystonia. They concluded that jaw-closing dystonia results in about 70% improvement, and that up to 50% of patients with jaw opening and lateralization dystonia, and lingual dystonia, benefit from BT injection, although dysphagia is a very common side effect for the latter two problems. A few subsequent studies have documented the effectiveness of BT injection in managing tongue protrusion dystonia and dysarthria associated with focal dystonia of orofacial or mandibular muscles. Thus, although further study is clearly necessary, BT injection seems to have potential as an effective treatment for lingual protrusion dystonia and orofacial and mandibular dystonias that affect speech.

Pharmacologic Management of Essential Voice Tremor

A small number of studies have addressed pharmacologic management (e.g., primidone, propranolol, methazolamide, clonazepam) of essential voice tremor or SD associated with essential voice tremor. In general, results suggest that pharmacologic management of SD and other hyperkinetic dysarthrias is not generally effective, although individual patients occasionally derive benefit. At this time, pharmacologic management cannot be considered a primary treatment for most individuals with hyperkinetic dysarthria.

Deep Brain Stimulation for Voice Tremor

One study (Taha, Janszen, & Favre, 1999). has reported the effect of bilateral thalamic deep brain stimulation (DBS) on voice tremor. Seven patients who had the surgery explicitly to manage their voice tremor were studied. Voice tremor was reportedly reduced by the DBS. Without considerable further study, however, DBS cannot be considered a primary treatment for voice tremor.

GENERAL CONCLUSION AND FUTURE RESEARCH NEEDS
A General Conclusion About Management for SD and Related Speech Conditions

The National Institutes of Health Consensus Development Conference on Clinical Use of Botulinum Toxin recommended that management be undertaken by “committed interdisciplinary teams” (American Academy of Neurology Therapeutics and Technology Assessment Subcommittee, 1990). The review undertaken for this report makes it very clear that the medical management of SD and related hyperkinetic speech disturbances, regardless of the specific treatment (e.g., botox injection, surgery), very often involves contributions from otolaryngologists, neurologists, and speech-language pathologists, and that, in general, the quality of published evidence is generally greatest when there has been collaboration among those subspecialties (and others, in some cases). Speech-language pathologists play several important roles in this context. These include (1) a crucial contribution to differential diagnosis, such as distinguishing SD from other dysarthrias, psychogenic or nonorganic dysphonia, and laryngeal muscular tension dysphonia, (2) perceptual and instrumental assessment of various aspects of speech and voice that are critical to measurement of baseline status, outcome, and subsequent modifications of treatment, and (3) patient counseling in all cases and voice treatment in selected cases. Based on the authorship characteristics in a substantial percentage of the published treatment literature for SD and related speech and voice disturbances that have been reviewed here, it appears that the interests of affected people are best served through collaboration among medical subspecialists and medical speech pathologists.

Future Research Needs

The following is a discussion of future research needs pertinent to intervention for SD and related disorders.

Treatment Comparisons

A variety of BT injections varying in terms of dose, site, and technique have been proposed. Most of these treatment comparisons utilized non-randomized, retrospective studies of clinical populations. If an accurate comparison between injection methods is a research goal, prospective randomized studies are needed.

Better Description of SD Patients

Individuals with SD form a somewhat heterogeneous group, yet frequently the only information about the participants in intervention studies is the fact that they have been diagnosed with SD. A standard set of criteria for the clinical diagnosis of SD would be of significant value for clinicians and researchers alike. More information about participants would allow the development of candidacy profiles. Such information would include the severity of SD (or severity of spasm), whether the dystonia is observed in multiple sites or restricted to the laryngeal area, familial history, presence of tremor, presence of pre-existing
reflux or swallowing problems, history of previous interventions and so on. Methods of better
differential diagnosis of SD from other disorders including motor neuron disease or essential
tremor are also needed because BT intervention may be inappropriate or ineffective for other
disorders.

Better Outcome Measures

This review indicates that there are many possible outcome measures, including patient self-
ratings and perceptual, aerodynamic, acoustic, and stroboscopic measures of vocal function.
Attempts at development of standard evaluation protocols for symptoms and severity of SD
have begun (Stewart et al., 1997) and should continue. If a parsimonious set of outcome
measures is to be developed, relationships among the various measures should be explored.
Of particular interest is the identification of those measures that are strongly related to vocal
quality. In addition, examination of the relationship among the variety of outcome measures
and the severity of the disorder is needed. Finally, outcome measures reflecting the subjects’
perception of vocal effort or speech fatigue are needed. Most of the measures used to reflect
treatment outcome focus on the level of the impairment. A better understanding of
psychosocial factors is warranted. These include studies of the effects of intervention on
employment, social participation, and quality of life. The focus on psychosocial issues is
particularly critical because voice quality impairment may not correlate well with measures of
limitations in activity and participation (Ma & Yiu, 2001).

Effect of Speech Treatment in Conjunction with BT Intervention

This review did not examine data regarding the effectiveness of behavioral intervention
(speech/voice therapy) for SD and related hyperkinetic speech disturbances, although the
literature search that was conducted suggests that such data are very limited and mostly
observational or testimonial. The literature on medical interventions for SD is very consistent
in indicating that voice therapy alone is not an effective intervention for SD. However, the
question remains whether speech intervention in conjunction with BT intervention would be
beneficial in extending the period between injections or allowing a lower injection dose (see
Murry and Woodson (1995) for relevant data). Closely associated with speech intervention is
the issue of compensation. How is the speaker compensating for the presence of SD? How
does this compensation change as a result of the sudden reduction in spasm associated with
BT intervention? What is the role of the respiratory system in these compensations? These
are questions that, if answered, would further define the nature of SD as well as the role of
behavioral intervention by speech-language pathologists in its management.

Understanding the Underlying Mechanism of BT Intervention

Understanding more thoroughly the mechanism underlying the functional effects of BT would
allow for the development of more appropriate types and timing of intervention. If the effects
are not limited to the neuromuscular junction, what central mechanism is involved? What are the physiologic bases for speech improvement?

**Long Term Effects**

The long terms effectiveness of BT for SD needs to be established. For example, how frequently do patients develop antibodies to BT and stop responding to injections? Do changes in injected muscles occur over time which alter the effectiveness of injection? These studies will become important as there is now a cohort of patients who have had BT treatment for more than a decade.

**Other Applications of BT**

BT intervention has been studied extensively in SD (especially the adductor type). More studies of BT in abductor SD are warranted, particularly with a focus on candidacy issues. Studies of other applications of BT intervention are also warranted including those for palatal myoclonus, voice tremor, swallowing difficulty as a result of cricopharyngeal spasm, and uncontrolled drooling (salivary duct injections).

**Alternatives to BT Intervention**

Further study of the effectiveness of alternatives to BT treatments are warranted. These alternatives include various surgical procedures designed to circumvent the drawbacks of RLN section and provide more lasting results than BT treatment.

**REFERENCES**

**General References**


**RLN Section references**


Botox Treatment References


**Miscellaneous References**


References Not Reviewed


*Revised by JRDuffy 11/3/01, KMY 11/6/01*