Critical Review:

which contributed to the purpose of this article. Reference lists were examined from articles retrieved for any further articles which contributed to the purpose of this review.

Selection Criteria

Studies included in this critical review were required to report speech outcomes following neurosurgical treatment for generalized dystonia. Studies examining outcomes for both primary and secondary dystonia were included. Studies that did not report results exclusive to participants with generalized dystonia (i.e., included segmental or focal dystonias in all reported outcomes) were not included.

Data Collection

Results of the literature search yielded papers on two different methods of surgical intervention. Papers from 1977-2001 described results of brain lesioning procedures done on the thalamus and globus pallidus. There were three papers on this topic reporting results of single-group post-test only (2), and single-group pre-posttest (1) study designs. and mixed (between and within) randomized clinical trial (1). Papers from 2005-2007 reported results on deep-brain stimulation of the globus pallidus. Study designs included single-group pre-posttest (2) and mixed (between and within) randomized clinical trial (1).

Results

Results of brain lesioning

Early studies on brain lesioning techniques report negative speech outcomes. Cooper (1977) performed bilateral or unilateral lesions to the ventrolateral nucleus and centrum medianum of the thalamus on 227 patients with primary and secondary generalized dystonia. After a 2-20 (mean 7.9) year follow-up interview was completed, the most commonly reported side effect was dysphonia, which occurred in 18% of patients who had bilateral lesions (n=122). While this study does include a large sample size, these results should be interpreted with caution due to lack of standardized follow-up times and absence of any statistical comparisons

Dysarthria was also found to be a common side effect for those undergoing thalamotomy procedures. Tasker, Doorly & Yamshiro (1988) reported results for 56 patients with primary and secondary dystonia who underwent unilateral and bilateral thalamotomy. A follow-up was completed with each available patient at the time of the study where, among other functions, dysarthria severity was rated by a neurologist on a scale of 0-5. Follow-up times ranged from less than 1 year to greater than 10 years after surgery. They found dysarthria to be a problem following both unilateral and bilateral lesions to the thalamus and included separate results for patients with primary and patients with secondary dystonia. Ten patients with primary dystonia (n=20) had post-operative difficulties with speech, which persisted at follow-up. Seven of these patients had undergone bilateral surgery. Transient dysarthria was observed in four patients with primary dystonia. In patients with secondary dystonia (n=30), four experienced worsened dysarthria, all of whom had undergone a bilateral procedure. Overall, any speech improvements were classified as “minimal.” Similar to Cooper (1977), the results of this study should be interpreted cautiously due to its lack of standardized follow-up times and statistical comparisons.

Better speech results were obtained for patients who received surgery targeting the globus pallidus as evidenced by Lin, Lin, Lin, Chang & Lee (2001). Their study included 18 patients with secondary generalized dystonia who received bilateral lesions. The Burke-Fahn-Marsden Dystonia Rating Scale (BFMDRS) is a validated tool commonly used to rate movement in people with dystonia (Burke et al. 1985). It includes a movement scale (based on a motor exam) and a disability scale (based on patient interview) on which to quantify the effects of dystonia (Ostrem & Starr, 2008). Within the movement scale, there is a rating for speech/swallowing movement. Within the disability scale, there is a rating for speech. The BFMDRS was

the activated and deactivated conditions. These same findings were maintained at 12 months post-operatively wh

(Burke et al., 1985), studies that use this scale are easier to compare. One difficulty is that none of the studies included in this review reported interrater or intrarater reliability on any of the measurements. Since these rating are subjective, reporting on these factors would make results more compelling.

Levels of evidence

The level of evidence provided by each of these studies also varies. Study design and methodology should be considered when deciding how much weight should be given to the results of a study. The two early single-group post-test only studies on brain lesioning did not include any statistical analysis of their findings (Cooper, 1977; Tasker et al., 1988) and therefore their findings cannot be deemed statistically significant. Cooper's study (1977) does not include the sex or age characteristics of his participants. His methods are also not detailed as he mentions that each patient had between 1 and 7 surgeries, with no breakdown of results according to extent of lesioning. Tasker's group (1988) also excludes some important information. They lost nine participants to follow-up, five of which were "surgical failures" and reasons for the remaining four are unreported. This missing data may have had an impact on results. The nature of these study designs (single-group post-test only) also neglects to report any information on patient functioning before surgical intervention.

As standards for research have improved with time, so has the evidence they present. Lin et al. (2001) produce more compelling evidence since they present their results using valid statistical analysis. However, their selection criteria lessen the strength of their findings. The authors selected the first 18 patients to reach a 12-month follow-up to include in their study. Those who did not reach the one-year follow-up or those who were unavailable may have had less favourable results. In contrast to the earlier studies on brain lesioning, Lin et al. (2001) found improved speech outcomes. Although they did use a different target site, these findings along with their selection criteria make the evidence from this study less compelling than the more recent studies on DBS. Their findings should also be considered with caution due to their small sample size.

One encouraging observation is that the more recent studies, specifically those examining DBS produce more compelling evidence with their results due to careful study design and detailed methodology. The three studies reporting results of DBS present very compelling results. All three used appropriate statistical tests (Wilcoxon signed-rank test) to determine significance of results. Kupsch's group (2006) used a control group for comparison and Vidailhet et al. (2005, 2007) used each subject as their own control, comparing all results to pre-intervention baseline measures. Missing data was accounted for in all three papers. Overall the three studies on DBS used experimental methods and could be replicated due to careful detailing of their procedures. Level of evidence from these papers could be improved with an increase in sample size.

Articles included in this critical review inadvertently follow the evolution of neurosurgical techniques for generalized dystonia. They begin with brain lesioning techniques targeting the thalamus, moving to similar procedures in the globus pallidus and finally the current preferred method of DBS in the globus pallidus. We can assume that as surgical techniques are refined, they have evolved to provide more control over dystonic movements of the body. The results of this critical review show that favourable results for speech are not as reliable. Early reports of brain lesioning studies reported that surgery to the thalamus had a negative impact on speech (Cooper, 1977; Tasker et al., 1988). One study reported improved speech after similar surgery with a different target site, the globus pallidus (Lin et al., 2001). Current studies on DBS report no change in speech (Vidailhet et al., 2005; Vidailhet et al., 2007) or some improvements with the potential for transient dysarthria (Kupsch et al., 2005). These results are congruent with other findings that point towards speech being controlled differently than other muscles