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Swallowing therapy – a prospective study on patients with neurogenic dysphagia due to unilateral paresis of the vagal nerve, Avellis' syndrome, Wallenberg's syndrome, posterior fossa tumours and cerebellar hemorrhage

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Summary

Background. No studies exist dealing with the outcome of dysphagic patients with posterior fossa (IV. ventricle) tumours (PFT) or cerebellar hemorrhage (CH), and the outcome of patients with Wallenberg's syndrome (WS) after functional swallowing therapy (FST) has so far not been studied in detail.

Patients and methods. 208 patients with neurogenic dysphagia (ND) who were consecutively admitted for functional swallowing therapy (FST) over a 3 year period to our hospital were examined clinically, by use of a videofluoroscopic swallowing study (VFSS) and/or fibreoptic evaluation of swallowing (FEES). The most frequent etiology was stroke (48%), followed by CNS tumours (13%). In the present study we defined three groups. Group 1 comprised 8 patients with PFT or CH. Group 2 consisted of 27 patients with WS, which was the leading cause among patients with non-hemispheric stroke. Since in WS a vagal nerve paresis due to affection of the Nucleus ambiguus occurs, 8 patients with Avellis' syndrome or unilateral paresis of the vagal nerve served as controls and were defined as group 3.

Findings. In the three groups, functional feeding status showed significant improvement after FST comprising methods of restitution, compensation and adaptation, each of which were applied in more than 80% of patients. Outcome was, however, significantly worse in group 1 as compared to group 2 and in group 2 as compared to group 3. Dysfunction of the upper esophageal sphincter and reflex triggering were significantly more severely disturbed in groups 1 and 2 as compared to group 3. Group 1 showed significantly more severe disturbances of the oral phase as compared to groups 2 and 3. After FST, more than 50% (5/8) of group 1 and 30% (8/27) of WS patients (group 2) were dependent on tube feeding, whereas all patients of group 3 were full-oral feeders.

Interpretation. This is the first study dealing with the outcome of dysphagic patients with PFT or CH. Based on our results it can be assumed that in these patients pressure is exerted on both dorsomedial central pattern generators (DMCPGs) for swallowing in a posterior-anterior direction. Due to the importance of the DMCPGs for swallowing, bilateral (and often MRI-invisible) lesions seem to be very harmful. For a better understanding of the pathomechanism responsible for ND in patients with PFT or CH, modern imaging methods such as proton magnetic resonance spectroscopy should be used for studying metabolic changes in the dorsal medulla in the future. Since the outcome of patients with WS with regard to dependence of tube feeding was not associated with the site or size of the lesion, it may – due to the individual asymmetry of the swallowing-dominant forebrain hemisphere – depend on the side of the medullary infarction.

Keywords: Dysphagia; outcome; Wallenberg's syndrome; posterior fossa tumour; cerebellar hemorrhage; Avellis' syndrome; central pattern generators for swallowing.

Introduction

Recently, we published the results of a prospective study on 208 patients with neurogenic dysphagia (ND) who were admitted to our hospital over a 3-year period for functional swallowing therapy (FST) [11]. The two most frequent etiologies in this population were stroke (48%) and brain tumours (13%). Whereas many studies exist which deal with hemisperic stroke and ND [3], ND has not been fully understood in patients with Wallenberg's syndrome (WS) and in those with posterior fossa (IV. ventricle) tumours (PFT). This is reflected by the fact that reports on the frequency of ND in WS vary in the literature between 51% and 100% [12] and that no study exists which deals with the association of ND and PFT. Since in WS, a vagal nerve paresis due to affection of the Nucleus ambiguus occurs, we chose patients with vagal nerve paresis due to Avellis' syndrome (AS) or unilateral peripheral paresis of the vagal nerve (PVN) as controls. In the presence of PFT, pressure is exerted to the brainstem in a posterior-anterior direction. Therefore, we also included patients with cerebellar hemorrhages (CH) because of a similar pathomechanism. In this article, we report on dysphagic patients with these etiologies.

Table 1. *Sample characteristics of the three patient groups and statistically significant differences within or between the groups after FST. Mean or median, range and standard deviation (SD) are shown (for abbreviations: see text); >denotes significantly better*

Patient groups	Age (mean, range, SD) years	Sex M : W	Duration of disease (median, range, SD) days	Duration of FST (median, range, SD) days	Number of patients dependent on tube feeding (TF) or tracheal cannula (TC)	FFS (comparison before and after FST)
Group 1 (n = 8)	57.9	3 : 5	54	72.5	Admission:	
– Posterior fossa tumours (n = 5)	(47–65)		(12–540)	(28–203)	TF: 8/8	
ependymoma °II (3)	7.1		182.2	70.6	TC: 7/8	
meningioma °I (1)						>
haemangioblastoma °1(1)					Discharge:	
– Cerebellar hemorrhage (n = 3)					TF: 5/8	
					TC: 4/8	
Group 2 (n = 27)	65.6	22 : 5	42	70	Admission:	
– Wallenberg's syndrome	(52–86)		(7–3500)	(4–210)	TF: 27/27	>
	9.9		679.5	51.6	Discharge:	
					TF: 8/27	
Group 3 (n = 8)	53.6	6 : 2	18.0	36.5	Admission:	
– Avellis' syndrome (n = 3)	(24–83)		(10–433)	(13–59)	TF: 5/8	
– Unilateral PVN (n = 5)	18.4		145.9	49	Discharge:	>
carotid endarterectomy (4)					TF: 0/8	
vagal neurinoma (1)						

Significant differences between groups after FST (combination of methods of restitution, compensation and adaptation in 80% of all patients): *FFS* group 3 > group 2; group 2 > group 1; *Disturbed reflex triggering* group 3 > groups 1, 2; *UES dysfunction* group 3 > groups 1, 2; *Oral phase problems* groups 2, 3 > group 1.

Patients and methods

For assessing the degree of ND on the activitation limitation level, as defined by the ICF of the WHO [18], we used an ordinal scale reflecting the functional feeding status as target variable (FFS) which was developed by us for this purpose [11]. VFSS and FFS evaluation before and after swallowing therapy were performed following standard protocols [5, 13]. The degree of penetration/aspiration was measured by use of 4-point ordinal scales developed by Schröter-Morasch [13] for FEES (from 1 = occasional aspiration, normal cough reflex to 4 = permanent aspiration, absent cough reflex, no sufficient volitional effort to eject) and by Hannig [5] for VFSS (from 1 = penetration to 4 = aspiration of >10% of the bolus volume and absent cough reflex). For the evaluation of a delayed swallowing reflex and amount of dysfunction of the upper esophageal sphincter (UES) we used the VFSS results (0 = normal, 1 = delayed/disturbed, 2 = severely disturbed/delayed/absent). Retentions in the valleculae and/or piriform sinuses were evaluated with regard to their side (unilateral, bilateral) and amount (0 = no retentions; 1 = mild/moderate retentions; 2 = massive retentions) by use of VFSS and/or FEES. Besides clinical, VFSS and FEES examinations an MRI was performed following a standard protocol on a Siemens Magnetom Vision 1.5 Tesla.

Wallenberg's syndrome [17] was diagnosed when dysphagic patients showed the triad of ipsilateral Horner's syndrome, ipsilateral ataxia and contralateral hypalgesia [12] in the presence of a dorsolateral infarction of the medulla oblongata. Avellis' syndrome was diagnosed according to the criteria as described elsewhere [1, 14]. The brainstem atlas of Olszewski and Baxter [9] was used for the identification of brainstem nuclei and pathways. Nonparametric tests were applied (Kruskal-Wallis test, Mann-Whitney U test, Wilcoxon test); statistical significance was set at $p < 0.05$ and corrected according to the Bonferroni procedure.

Results

Sample characteristics and significant differences within and between patient groups are shown in Table 1.

Discussion

Although functional outcome showed a significant improvement after FST in each of the three groups, there were significant differences between them. Patients with WS had a better functional outcome as compared to patients with PFT or CH and a worse outcome as compared to patients with AS or PVN. 30% of WS patients were dependent on tube feeding after FST. This high percentage is in agreement with the results of other studies on WS patients who were admitted to a rehabilitation service. E.g., in the study of Meng *et al.* [6], 22% of patients with brainstem stroke remained dependent on tube feeding; in the study of Teasell *et al.* [15] even four of seven WS patients bearing a PEG (out of a total of 11 patients) could not resume oral intake at discharge. 18 of 27 patients with WS (66.7%) had bilateral pharyngeal retentions, which was described earlier by our research group [8] and other authors [2, 16] and may be ex-

plained by a disconnection syndrome between the ipsilateral pattern generators for swallowing and the contralateral side of the medulla, as proposed by Aydogdu *et al.* [2]. WS patients as well as those with PFT or CH had significantly more severe disturbances of UES opening and reflex triggering as compared to patients with AS or PVP. Therefore, besides FST, cricopharyngeal myotomy or botulinum toxin injection into the UES have to be considered in patients with WS, PFT or CH, especially in cases of severe and FST-resistant UES dysfunction as revealed by use of manometry. Finally, patients with PFT or CH showed a significantly more severely disturbed oral phase as compared to WS and AS patients.

It is worth mentioning that all WS patients had their infarctions in the most rostral part of the medulla. Our results point to the fact that the dorsomedial central pattern generators (DMCPGs) for swallowing, which are situated near the Nucleus tractus solitarii [7] and are affected in WS, are more important with regard to swallowing functions than the ventrolateral central pattern generators (VLCPGs), which lie near the Nucleus ambiguus and are affected, e.g., in patients with Avellis' syndrome. Since the outcome of patients with WS, with regard to dependence of tube feeding, was not associated with their lesion size or site, it may – due to the individual asymmetry of the swallowing-dominant forebrain hemisphere [4] – depend on the side of their medullary infarction.

As far as we know, this is the first study dealing with the outcome of dysphagic patients with PFT or CH. In the study of Perie *et al.* [10], only one patient with a longlasting ND due to CH was described. Based on our results it can be assumed that in these patients pressure is exerted on both DMCPGs in a posterior-anterior direction. Due to the importance of the DMCPGs for swallowing, bilateral (and often invisible) lesions seem to be very harmful. Hypothetically, the most severe ND would be found in patients with bilateral lesions of both DMCPGs and VLCPGs. Probably, such a "bilateral Wallenberg's syndrome" cannot be survived. For a better understanding of the pathomechanism responsible for ND in patients with PFT or CH, modern imaging methods such as proton magnetic resonance spectroscopy should be used for studying metabolic changes in the dorsal medulla in the future.

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