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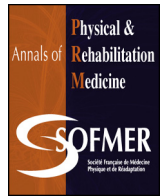
Original article

Swallowing in individuals with disorders of consciousness: A cohort study

Evelyne Mélotte [a,b,c,*], Audrey Maudoux [d,e], Sabrina Delhalle [e], Aude Lagier [e], Aurore Thibaut [b,c], Charlène Aubinet [b,c], Jean-François Kaux [a], Audrey Vanhaudenhuyse [d,f], Didier Ledoux [g,1], Steven Laureys [b,c,1], Olivia Gosseries [b,c,1]

[a] Physical and Rehabilitation Medicine Department, University Hospital of Liege, Liege, Belgium
[b] Coma Science Group, GIGA-Consciousness, University of Liege, Liege, Belgium
[c] Centre du Cerveau², University Hospital of Liege, Liège, Belgium
[d] Sensation and Perception Research Group GIGA, University of Liege, Liege, Belgium
[e] Otorhinolaryngology Head and Neck Surgery Department, University Hospital of Liege, Liege, Belgium
[f] Algology Department, University Hospital of Liege, Liege, Belgium
[g] Intensive Care Unit Department, University Hospital of Liege, Liege, Belgium

ARTICLE INFO

ABSTRACT

Background: After a period of coma, a proportion of individuals with severe brain injury remain in an altered state of consciousness before regaining partial or complete recovery. Individuals with disorders of consciousness (DOC) classically receive hydration and nutrition through an enteral-feeding tube. However, the real impact of the level of consciousness on an individual's swallowing ability remains poorly investigated.

Objective: We aimed to document the incidence and characteristics of dysphagia in DOC individuals and to evaluate the link between different components of swallowing and the level of consciousness.

Methods: We analyzed clinical data on the respiratory status, oral feeding and otolaryngologic examination of swallowing in DOC individuals. We analyzed the association of components of swallowing and participant groups (i.e., unresponsive wakefulness syndrome [UWS] and minimally conscious state [MCS]).

Results: We included 92 individuals with DOC (26 UWS and 66 MCS). Overall, 99% of the participants showed deficits in the oral and/or pharyngeal phase of swallowing. As compared with the MCS group, the UWS group more frequently had a tracheostomy (69% vs 24%), with diminished cough reflex (27% vs 54%) and no effective oral phase (0% vs 21%).

Conclusion: Almost all DOC participants had severe dysphagia. Some components of swallowing (i.e., tracheostomy, cough reflex and efficacy of the oral phase of swallowing) were related to consciousness. In particular, no UWS participant had an efficient oral phase, which suggests that its presence may be a sign of consciousness. In addition, no UWS participant could be fed entirely orally, whereas no MCS participant orally received ordinary food. Our study also confirms that objective swallowing assessment can be successfully completed in DOC individuals and that specific care is needed to treat severe dysphagia in DOC.

1. Introduction

After a period of coma, individuals with severe brain injury may remain in an altered state of consciousness before regaining partial or complete recovery [1]. Disorders of consciousness (DOC) consist of 3 states ranging from no awareness and no arousal to the preservation of arousal with fluctuating awareness [2]: coma, vegetative state/unresponsive wakefulness syndrome (UWS) [3] and minimally conscious state (MCS) [4]. UWS is characterized by the presence of eye-opening and reflexive movements, without conscious behaviours [3]. Individuals with MCS show reproducible but inconsistent signs of consciousness, such as command following, visual pursuit, and localization to noxious stimulation

[4]. When they recover the ability to functionally communicate or to use objects adequately, they emerge from the MCS [4].

Swallowing disorders are relatively frequent after an acquired brain injury from traumatic or anoxic causes, ranging from 25% to 61% depending on the studies [5,6]. Oral feeding has been suggested to be related to the level of consciousness in previous studies evaluating swallowing in severe brain-injured individuals [5–17]. Individuals with DOC classically receive hydration and nutrition through an enteral-feeding tube [18]. The real impact of the level of consciousness on individuals' swallowing ability remains poorly investigated. Indeed, previous studies that assessed swallowing in individuals with severe brain injury had small sample sizes [15], focused on only one component such as the type of feeding [17] or the possibility of extubation [16] and/or they mostly used clinical assessment of consciousness that was not based on established diagnostic criteria [4] (see [5–14]). In line with this, previous literature showed that the Coma Recovery Scale-Revised (CRS-R) [19] is the current gold standard to assess the level of consciousness in DOC individuals, and multiple evaluations should be performed to decrease misdiagnosis [20]. Accurate diagnosis is challenging because of confounding factors such as aphasia and motor deficits, but it has important implications for prognosis [21], treatment management [22] and related ethical considerations [23].

Swallowing has not yet been studied systematically in DOC individuals, and to our knowledge, the link between the level of consciousness and swallowing components (e.g., lip prehension, lingual propulsion, pharyngo-laryngeal sensitivity, efficacy of the pharyngeal phase and ability to clear saliva) has never been investigated. We recently suggested that an effective oral phase of swallowing could be a determinant to consider swallowing as a conscious behavior [17]. However, in this previous study, we included only UWS participants and based our conclusion on only 2 components (i.e., presence or absence of oral feeding and type of oral feeding).

In the present work, we collected respiratory and nutritional status as well as the otolaryngological results of swallowing in a large cohort of individuals with prolonged DOC. We predicted that most individuals have severe alterations of the different components of swallowing and that some components of the swallowing process, such as the oral phase, may be linked to the level of consciousness.

2. Participants and methods

2.1. Participants

We retrospectively collected data for individuals admitted consecutively from January 2010 to August 2018 to the University Hospital of Liege (Belgium) for a 1-week multimodal assessment of consciousness for diagnostic and prognostic purposes. Fig. 1 illustrates the flow of participants in the study. Inclusion criteria were 1) recovered from coma caused by a severe acquired brain injury, 2) with a prolonged condition (> 1 month post-insult) [25,26], 3) medically stable, 4) underwent fiberoptic endoscopic examination of swallowing (FEES), 5) a diagnosis of UWS or MCS based on a minimum of 5 CRS-R tests (to avoid diagnostic errors due to fluctuations in responsiveness and to obtain a stable clinical
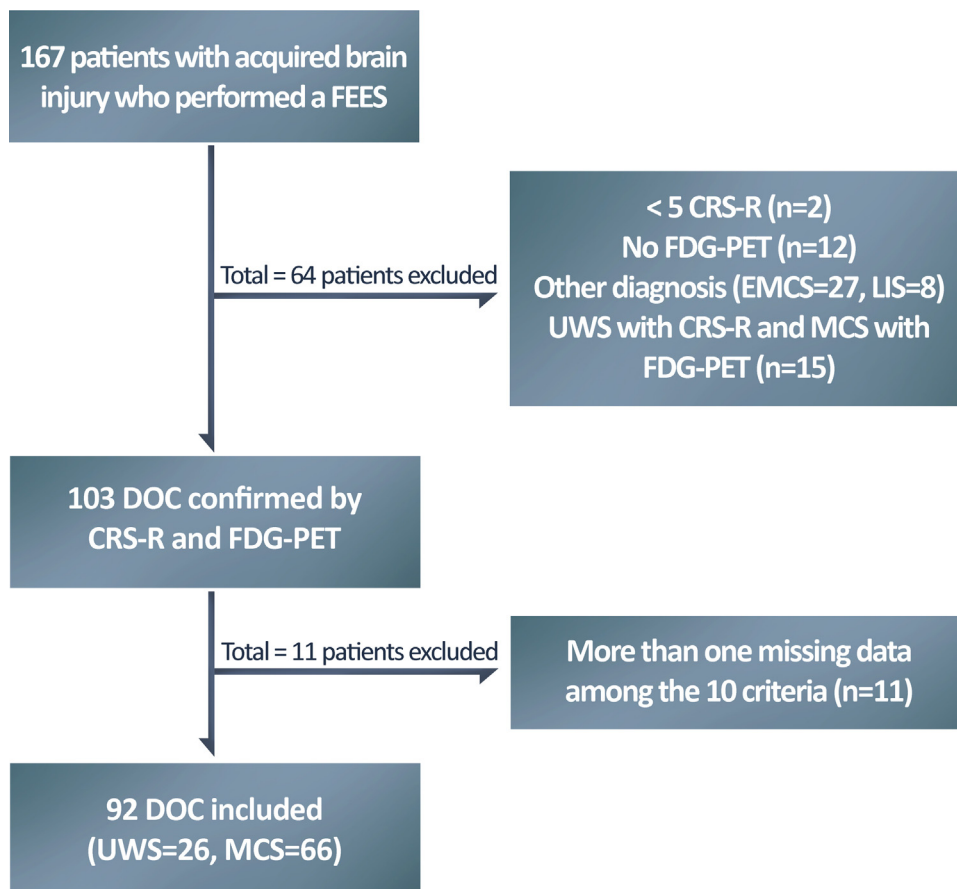

Fig. 1. Flowchart of the study. FEES: fiberoptic endoscopic evaluation of swallowing; CRS-R: Coma Recovery Scale-Revised; FDG-PET: fluorodeoxyglucose-positron emission tomography; EMCS: emergence from minimally conscious state; LIS: locked-in syndrome; DOC: disorders of consciousness; UWS: unresponsive wakefulness syndrome; MCS: minimally conscious state.

diagnosis) [20], which was confirmed on fluorodeoxyglucose-positron emission tomography (FDG-PET) (see next sections concerning data acquisition and analyses) and 6) no more than 1 missing data item (i.e., at least 9 of 10 selected criteria regarding respiratory status, type of feeding and components of the oral and pharyngeal phases of swallowing).

We extracted demographic data (i.e., sex, age, etiology, time since insult) and DOC diagnosis from participants' medical records. Etiology was classified in terms of focal or global injury to distinguish between on one hand, ischemic, hemorrhagic and traumatic brain injury, and on the other, anoxic and metabolic encephalopathy.

The study was approved by the Ethics Committee of the Faculty of Medicine of the University Hospital of Liege, and written informed consent was obtained from all legal surrogates. We followed the principles of the STrengthening the Reporting of OBservational studies in Epidemiology (STROBE) (see Supplementary materials) [24].

2.2. Diagnosis of consciousness

The diagnosis was established after repeated behavioral assessments performed by trained and experienced clinician researchers using the CRS-R [19] and FDG-PET. The CRS-R consists of 6 subscales (auditory, visual, motor, and oromotor functions as well as communication and arousal) of 23 items ordered by degree of complexity, ranging from reflexive to cognitively mediated behaviors [27]. All participants underwent at least 5 CRS-R tests over a maximum of 10 days and the best result was kept for the behavioral diagnosis [20]. To confirm the behavioral diagnosis, FDG-PET images were visually inspected by experts and classified as compatible with UWS (when the statistical tool detected no voxels with preserved metabolism in the associative fronto-parietal network bilaterally) or compatible with MCS (with incomplete hypometabolism or partial preservation of metabolic activity detected in the fronto-parietal network). The acquisition procedure and analyses of FDG-PET data were described previously [28,29]. The brain activity map was obtained with a threshold of uncorrected $p < 0.05$ in all contrasts for single-subject analyses, as in previous studies [28,29]. We excluded individuals with a diagnosis of UWS based on the CRS-R but with FDG-PET results compatible with MCS because these individuals may present covert consciousness (referred to as MCS*) [2].

2.3. Respiratory status, type of feeding and swallowing assessments

We collected data on 10 specific criteria based on the results of the ear, nose and throat (ENT) examination (performed by SD, AM or AL) and from the questionnaires completed by the family (for the type of feeding).

For respiratory status, we reported the presence or absence of tracheostomy (criterion 1). The type of feeding (criterion 2) referred to the presence or absence of exclusive enteral-feeding. For participants who received oral feeding, we distinguished type of feeding based on the criteria of the Food Intake Level Scale (FILS) [30]. The FILS is an observer-rating scale for assessing the severity of dysphagia, examining to what degree individuals take food orally on a daily basis, ranging from 0 (no oral intake, and no swallowing training) to 10 (normal oral food intake). Scores 1 to 3 correspond to no oral intake, 4 to 6 oral intake with alternative nutrition, and 7 to 10 oral intake exclusively.

The other 8 criteria were related to the otolaryngological examination performed by ENT experts. A FEES was performed with a flexible videorhinolaryngoscope (Olympus Visera OTP-S7, Tokyo) and color monitor. We excluded all criteria that required a response to a command (e.g., assessment of the nasopharyngeal or

vocal fold closure with the production of sounds, apnea, volunteer saliva swallow, cough). The first 3 criteria of the otolaryngological examination were related to the oral phase of swallowing with the presence or absence of hypertonia of jaw muscles (criterion 3), the presence or absence of an oral phase of swallowing (lip prehension or lingual propulsion; criterion 4), and the observation or not of an effective oral phase (criterion 5). Practically, we moved a spoon to in front of the individual's mouth and observed the reaction. With absence of lip prehension, we placed a 2-ml bolus in the middle of the tongue and observed if lingual propulsion occurred. We considered the oral phase effective if we detected consecutively lip prehension, lingual propulsion and no post-swallowing oral stasis. The last 4 criteria were related to the pharyngeal phase of swallowing. The presence or absence of secretions in the pharyngo-laryngeal area (criterion 6) and the salivary aspiration (criterion 7) informed on participants' ability to manage secretions. The cough reflex (criterion 8) was evaluated by stimulating the laryngeal area, and if no cough was observed, the pharyngo-laryngeal sensitivity was considered absent. Finally, we noted the presence or absence of bolus aspiration during the swallowing of 2 ml of thick and liquid textures (criteria 9 and 10, respectively). Some participants did not undergo the functional swallowing test because of a severe bite reflex, an inefficacity of the oral phase, or because it was considered too dangerous regarding other parameters (e.g., too many saliva aspirations, absence of spontaneous saliva swallowing).

2.4. Statistical analysis

We performed a descriptive analysis for each diagnosis group (UWS and MCS) in terms of sex, age, etiology and time since insult. Normality was assessed with histograms, quantile plots and Shapiro-Wilk tests. Univariate comparisons between UWS and MCS groups involved chi-square or Fisher exact test for categorical variables and Kruskal-Wallis test for non-normally distributed continuous variables. The association between each of the 10 criteria and the diagnosis groups was assessed by univariate logistic regression. These associations were further investigated by multivariable logistic regression adjusted for etiology and time since insult. The results of logistic regressions are presented as odds ratios (ORs) and adjusted ORs together with their 95% confidence intervals (CIs). $P < 0.05$ was considered statistically significant. Individuals with missing values were excluded from the analysis for the considered criteria. Statistical analyses were performed with Stata v14.2 (Stata Corp. 2015, College Station, TX).

3. Results

Among the 167 individuals identified, 92 matched our inclusion criteria (Fig. 1); 26 showed only reflexive behavior and had a diagnosis of UWS and 66 satisfied the CRS-R criteria for MCS in at least one evaluation (Table 1) Eleven of the 26 UWS participants were already included in our previous UWS studies focusing on type of feeding exclusively [17]. Diagnosis was confirmed by FDG-

Table 1
Descriptive statistics of the whole sample.

	UWS (n = 26)	MCS (n = 66)	P-value
Sex (F/M)	9/17	30/36	0.343
Age, years, mean (SD)	41 (12)	38 (12)	0.405
Etiology	Focal: 11	Focal: 49	0.004
	Global: 15	Global: 17	
Time since injury, months, mean (SD)	30 (22)	4 (34)	0.014

UWS: unresponsive wakefulness syndrome; MCS: minimally conscious state.

PET in all included participants. The two diagnosis groups did not differ in age or sex but did differ by etiology and time since insult (Table 1), MCS participants having longer time since insult and more focal damage than UWS participants. We first present the descriptive analysis (Table 1) and then the percentage of participants for each of the 10 selected criteria, the univariate analysis for each criterion by diagnosis (p_{uni}), and the adjusted multivariate analysis ($p_{adjusted}$) for etiology and time since insult (Table 2). We also illustrate the percentage of UWS and MCS participants for each criterion in Fig. 2.

In total, 91 of 92 participants presented disorders in at least one criterion linked to the oral or pharyngeal phase of swallowing (i.e., criteria 3 to 10). Regarding respiratory status, 34 (37%) participants still had a tracheostomy at the time of assessment. As compared with MCS individuals, UWS individuals more frequently had a tracheostomy ($p_{adjusted}$ = 0.002). In total, 71 (77%) participants received enteral feeding exclusively, with no significant difference between UWS and MCS groups ($p_{adjusted}$ = 0.254). None of the MCS participants received exclusive ordinary solid food (FILS 10). Regarding the ENT examination, for the oral phase, 52 (56%) participants presented hypertonia of the jaw muscles, with no difference between UWS and MCS groups ($p_{adjusted}$ = 0.881). In total, 43 (47%) participants showed at least one component of the oral phase of swallowing (lip prehension or lingual propulsion), with no difference between groups ($p_{adjusted}$ = 0.94). However, UWS and MCS groups differed in efficacy of the oral phase of swallowing ($p_{adjusted}$ = 0.011), characterized by the presence of lip prehension and lingual propulsion without post-swallowing oral stasis. For the pharyngeal phase, 34 (37%) participants had pharyngo-laryngeal secretions and 26 (28%) saliva aspiration. UWS and MCS groups differed on univariate analysis for the pharyngo-laryngeal secretions (p_{uni} = 0.012) and saliva aspiration (p_{uni} = 0.019) but not significantly on multivariable analysis (pharyngo-laryngeal secretions: $p_{adjusted}$ = 0.067; saliva aspiration: $p_{adjusted}$ = 0.062). For the test of the cough reflex, among the 63 participants assessed, 43 (52%) showed decreased pharyngo-laryngeal sensitivity, with significantly more MCS participants presenting a cough reflex than UWS participants ($p_{adjusted}$ = 0.027).

Regarding the functional test with thick and liquid texture, 16 (61%) and 53 (80%) UWS and MCS participants, respectively, performed the swallowing test with a cream texture and 12 (46%) and 42 (63%) with a liquid texture. Nine (13%) participants showed aspiration with a thick texture and 17 (31%) with a liquid texture, with no difference between groups (thick texture: $p_{adjusted}$ = 0.798; liquid texture: $p_{adjusted}$ = 0.226).

4. Discussion

The main aims of this study were to document the proportion and characteristics of dysphagia in individuals with DOC and to evaluate the link between different criteria of swallowing and level of consciousness. To our knowledge, this is the first study of respiratory status, oral feeding and FEES in a large cohort of individuals with DOC. In our study, all but one DOC individual (MCS) presented at least one swallowing dysfunction of the oral and/or pharyngeal phase. Also, tracheostomy, cough reflex and the efficacy of the oral phase were the 3 criteria related to consciousness. Finally, none of the UWS individuals could be fed entirely orally, whereas none of the MCS individuals received ordinary oral food.

Regarding type of feeding, none of the UWS participants could achieve a full oral feeding, most probably linked to the absence of an effective swallowing oral phase and less effective pharyngeal phase, as shown by the proportion of participants with tracheostomy. Only a small proportion (7%) of MCS participants could safely resume full oral feeding with easy-to-swallow food (i.e., FILS 7). Despite the ability of some MCS participants to resume oral feeding, a higher level of consciousness (i.e., EMCS) is probably necessary to enable a full ordinary oral food (FILS 10).

Some swallowing criteria were notably related to the level of consciousness. First, UWS and MCS participants differed in spontaneous saliva management because more UWS participants still had a tracheostomy at the time of the evaluation than MCS participants. None of the participants were respirator-dependent. The need for the tracheostomy in about one third of the participants can probably be explained by insufficient saliva

Table 2
Descriptive and statistical analysis of the 10 criteria in the UWS and MCS groups.

Criteria	UWS n (%)	MCS n (%)	p_{uni}	$p_{adjusted}$	UWS vs MCS OR	95% CI
1. Tracheostomy still in place	18/26 (69.2)	16/66 (24.2)	< 0.001	0.002	5.67	1.86–17.27
Tracheostomy removed	5	38				
Never had a tracheostomy	3	12				
2. Full enteral feeding	23/26 (88.5)	48/66 (72.7)	0.117	0.254	2.45	0.53–11.39
Partial oral feeding (FILS 7)	3	13				
Full oral feeding (FILS 7)	0	3				
Full oral feeding (FILS 7)	0	2				
Oral phase						
3. Hypertonia of the jaw muscles	15/26 (57.7)	37/66 (56.1)	0.887	0.881	1.08	0.41–2.87
4. Oral phase	12/26 (46.2)	31/66 (47.0)	0.844	0.94	0.96	0.37–2.53
5. Efficacy of the oral phase	0/26 (0)	14/66 (21.2)	0.007	0.011	0.09	0–0.63
Pharyngeal phase						
6. Pharyngo-laryngeal secretions	15/26 (57.7)	19/66 (28.8)	0.012	0.067	2.53	0.94–6.82
7. Saliva aspiration	12/26 (46.2)	14/66 (21.2)	0.019	0.062	2.65	0.95–7.38
8. Cough reflex	7/23 (30.4)	36/60 (60)	0.019	0.027	0.30	0.10–0.87
9. Cream aspiration	2/16 (12.5)	7/53 (13.2)	0.941	0.798	1.26	0.21–7.44
Not performed[a]	10	13				
10. Liquid aspiration	2/12 (16.7)	15/42 (35.7)	0.223	0.226	0.35	0.07–1.90
Not performed[a]	14	24				

p_{uni}: univariate analysis between UWS and MCS; $p_{adjusted}$: multivariable analysis between UWS and MCS adjusted for etiology and time since insult; PL: pharyngo-laryngeal; OR: odds ratio; CI: confidence interval; FILS: Food Intake Level Scale.
[a] Not performed because it was considered too risky by examiners or because of troubles in the oral phase (e.g., bite reflex, no lingual propulsion).
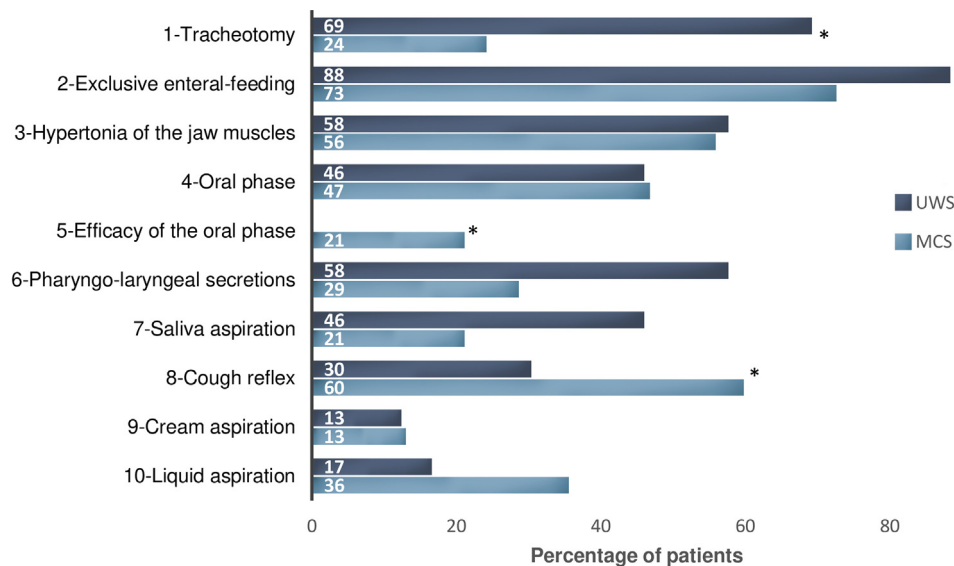
Fig. 2. Percentage of UWS and MCS patients for the 10 criteria. UWS: unresponsive wakefulness syndrome; MCS: minimally conscious state. Asterisks (*) indicate significant difference between UWS and MCS, P adjusted < 0.05.

swallowing reflexes and ineffective pharyngeal phase of swallowing. Also, we observed more pharyngo-laryngeal secretions and saliva aspiration in UWS than MCS participants although not significantly after controlling for time since injury and etiology. These findings suggest that the level of consciousness may affect the ability to correctly manage saliva.

Second, both UWS and MCS participants showed a partial oral phase of swallowing (e.g., tongue and masticatory-like movements). However, no UWS participants and only a small number of MCS participants showed an effective oral phase of swallowing (lip prehension, lingual propulsion and no post-swallowing oral stasis). From this result and as already suggested in our previous study [17], an effective oral phase should be considered a sign of consciousness and thus should be taken into account in DOC diagnosis. The oral phase of swallowing is usually described as the voluntary (conscious) part of swallowing, controlled by multiple cortical regions such as the primary sensori-motor cortex, supplementary motor area, premotor cortex (also called "cortical masticatory area"), thalamus, cingulate, putamen and insulo-opercular cortex that interact with regions in the brainstem [31–34]. Some authors showed that masticatory-like movements and rhythmic tongue activity can be produced by the recruitment of the brainstem alone [35,36]. However, although the brainstem controls basic activity patterns of the cranial motoneuron groups involved in the oral phase of swallowing (hypoglossal, trigeminal, facial, vagal), descending inputs from the central nervous system also play an important role. Indeed, for some authors [36], the activity pattern of each motoneuron is modulated by higher brain and peripheral inputs. This cortical and peripheral recruitment allows that the final motor outputs fit the environmental demand. If the components of the oral phase of swallowing (e.g., mastication) were based on only a central pattern generator in the brainstem, it would be stereotyped [37]. Thus, the presence of an effective oral phase of swallowing seems highly dependent on cortical recruitment, which would explain why UWS individuals who show severe alterations of supratentorial cerebral metabolism do not present an effective oral phase of swallowing.

Finally, the presence of a cough reflex was a criterion more present in MCS than UWS participants. As shown in neuroimaging studies [38], the cough reflex is probably not a simple pontomedullary reflex arc. Indeed, in these previous studies, the cough reflex was facilitated by cortical activations (mainly the primary motor cortex, posterior insula, paracentral lobule, posterior mid-cingulate cortex, premotor cortex). Thus, the impact of the level of consciousness highlighted in the present study might probably be linked to the importance of the underlying cortical damage. Moreover, it is now generally accepted in stroke literature that the cortex plays an important role in the control of swallowing and that damages to swallowing motor areas and/or their connection to the brainstem usually result in dysphagia [39,40].

Besides these main findings, hypertonia of the jaw muscles and the number of aspirations on cream/liquid texture did not differ between UWS and MCS groups. However, more MCS participants performed the functional test with thick and liquid texture than did UWS participants, which highlights that UWS participants seem to be generally considered more at risk of aspiration by the examiners or had more troubles in the oral phase (e.g., hypertonicity, absence of lip prehension or lingual propulsion) than MCS participants.

Our findings also agree with previous studies showing that objective swallowing assessment such as the FEES can be successfully completed in DOC individuals [7,9,15]. The ENT examination gives precious information on 2 main points, first regarding tracheostomy and second regarding the possibility of oral feeding. After the FEES, the ENT specialist suggested removing the tracheostomy for several DOC participants (8 MCS participants and 6 UWS participants) because of good saliva management and the absence of stenosis or laryngeal paralysis. In these cases, the tracheostomy was probably maintained to prevent any respiratory complications because of the lack of adequate information regarding the management of the tracheostomy or because no one tested the possibility to begin a tracheostomy weaning. A previous study reported that individuals with a tracheostomy are more likely to develop pneumonia than patients without a tracheostomy [41]. This should be kept in mind when making decisions on the need to maintain a tracheostomy in this fragile population. In this regard, the FEES can help in deciding on possible decannulation, but given the complexities of saliva management in the DOC population, the decision for tracheostomy weaning should be discussed in a multidisciplinary team. Besides tracheotomy management, the ENT examination is crucial for patients in whom we would like to start or continue oral feeding. Indeed, with the high proportion of patients with absence of cough reflex (about 54% in the whole sample), there is a high risk of silent aspiration. In

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this study, the ENT examination also advised starting partial oral feeding in 3 more UWS participants and 13 MCS participants and to stop full or partial oral feeding in 3 MCS participants. A functional swallowing test can be difficult to implement in individuals with severe trismus or total absence of an oral phase of swallowing, but as long as partial oral phase is present (e.g., partial lip prehension and lingual propulsion), thick or liquid swallowing can be tested by a qualified clinician.

Regarding the severity of dysphagia in DOC individuals, specific care such as nursing, chest physiotherapy, and speech therapy are recommended [42]. Management of swallowing should be integrated into a global approach taking into account respiration, mobility and tonicity of the face and considering emotional reactions, spasticity, and potential hypersensitivity. Clinically, we noticed that therapeutic feeding (i.e., swallowing small amounts of tasty easy-to-swallow food) can sometimes help clear excess saliva secretions in the pharyngo-laryngeal area. In addition, taste stimulation (involving only a very small amount of food or liquid that is delivered via a cotton swab in particular zones of the oral cavity) is also a good option for individuals who are at risk of aspiration. Nevertheless, the decision to introduce oral food or liquid in DOC individuals as therapeutic feeding or as a real part of the feeding should only be made after the completion of an objective swallowing evaluation [9].

There are several limitations of the study related to its retrospective, observational and single-centre design, which suggests that our data are moderately representative of the general DOC population. The presence of missing data for one criterion (cough reflex) should be acknowledged. We were also limited in the number of available criteria that could be studied. A prospective analysis, given a prescribed test protocol for DOC individuals with more detailed criteria, may yield additional information that were not covered in the current study, such as the duration of the oral phase before the swallowing reflex, the frequency of spontaneous saliva swallowing reflex [13] or the importance of secretions post-swallowing with a valid protocol such as the New Zealand secretion scale [43]. Future longitudinal studies should also investigate the recovery of dysphagia along with consciousness recovery within the same individuals.

In conclusion, this study provides promising results linking swallowing and consciousness, notably regarding the ability of (minimally) "conscious" individuals to better manage spontaneous swallowing of saliva (reflected here by the presence or absence of tracheostomy), the cough reflex and the efficiency of the oral phase of swallowing. We should continue our efforts in the assessment of the oro-facial area in DOC individuals to be able to propose appropriate and sensible care management.

Author contributions

E.M., O.G., and S.L. contributed to conception and design of the study. E.M., S.D., A.M., and A.L., acquired the ENT data. OG, A.T., C.A. and A.V. acquired the diagnosis data. E.M., OG and D.L. analyzed the data. E.M. and O.G. drafted the manuscript. A.M., A.L., A.T., C.A., JF.K., A.V., D.L., and S.L. provided major revisions in significant proportions of the manuscript. D.L., O.G. and S.L. contributed equally.

Disclosure of interest

The authors declare that they have no competing interest.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.rehab.2020.04.008.

References

[1] Schnakers C. Coma and disorders of consciousness, 2nd ed., New York, NY: Springer Berlin Heidelberg; 2017.

[2] Gosseries O, Zasler ND, Laureys S. Recent advances in disorders of consciousness: focus on the diagnosis. Brain Inj 2014;28:1141–50. http://dx.doi.org/10.3109/02699052.2014.920522.

[3] Laureys S, Celesia GG, Lavrijsen J, León-Carrión J, Sannita WG, Sazbon L, et al. Unresponsive wakefulness syndrome: a new name for the vegetative state or apallic syndrome. BMC Med 2010;1:68. http://dx.doi.org/10.1186/1741-7015-8-68.

[4] Giacino JT, Ashwal S, Childs N, Cranford R, Jennett B, Katz DI, et al. The minimally conscious state: definition and diagnostic criteria. Neurology 2002;58:349–53.

[5] Mackay LE, Morgan AS, Bernstein BA. Swallowing disorders in severe brain injury: risk factors affecting return to oral intake. Arch Phys Med Rehabil 1999;80:365–71. http://dx.doi.org/10.1016/S0003-9993(99)90271-X.

[6] Winstein CJ. Frequency, progression, and outcome in adults following head injury. Phys Ther 1983;63:8.

[7] O'Neil-Pirozzi TM, Jack Momose K, Mello J, Lepak P, McCabe M, Connors JJ, et al. Feasibility of swallowing interventions for tracheostomized individuals with severely disordered consciousness following traumatic brain injury. Brain Inj 2003;17:389–99. http://dx.doi.org/10.1080/0269905031000070251.

[8] Formisano R, Voogt RD, Buzzi MG, Vinicola V, Penta F, Peppe A, et al. Time interval of oral feeding recovery as a prognostic factor in severe traumatic brain injury. Brain Inj 2004;18:103–9. http://dx.doi.org/10.1080/0269905031000149470.

[9] Brady SL, Darragh M, Escobar NG, O'neil C, Pape TLB, Rao N, et al. Persons with disorders of consciousness: are oral feedings safe/effective? Brain Inj 2006;20:1329–34. http://dx.doi.org/10.1080/02699050601111435.

[10] Terré R, Mearin F. Prospective evaluation of oro-pharyngeal dysphagia after severe traumatic brain injury. Brain Inj 2007;21:1411–7. http://dx.doi.org/10.1080/02699050701785096.

[11] Hansen TS, Engberg AW, Larsen K. Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury. Arch Phys Med Rehabil 2008;89:1556–62. http://dx.doi.org/10.1016/j.apmr.2007.11.063.

[12] Terré R, Mearin F. Evolution of tracheal aspiration in severe traumatic brain injury-related oropharyngeal dysphagia: 1-year longitudinal follow-up study. Neurogastroenterol Motil 2009;21:361–9. http://dx.doi.org/10.1111/j.1365-2982.2008.01208.x.

[13] Crary MA, Sura L, Carnaby G. Validation and demonstration of an isolated acoustic recording technique to estimate spontaneous swallow frequency. Dysphagia 2013;28:86–94. http://dx.doi.org/10.1007/s00455-012-9416-y.

[14] Mandaville A, Ray A, Robertson H, Foster C, Jesser C. A retrospective review of swallow dysfunction in patients with severe traumatic brain injury. Dysphagia 2014;29:310–8. http://dx.doi.org/10.1007/s00455-013-9509-2.

[15] Bremare A, Rapin A, Veber B, Beuret-Blanquart F, Verin E. Swallowing disorders in severe brain injury in the arousal phase. Dysphagia 2016;31:511–20. http://dx.doi.org/10.1007/s00455-016-9707-9.

[16] Godet T, Chabanne R, Marin J, Kauffmann S, Futier E, Pereira B, et al. Extubation failure in brain-injured patients: risk factors and development of a prediction score in a preliminary prospective cohort study. Anesthesiology 2017;126:104–14. http://dx.doi.org/10.1097/ALN.0000000000001379.

[17] Mélotte E, Maudoux A, Delhalle S, Martial C, Antonopoulos G, Larroque SK, et al. Is oral feeding compatible with an unresponsive wakefulness syndrome? J Neurol 2018;265:954–61. http://dx.doi.org/10.1007/s00415-018-8794-y.

[18] Whyte J, Laborde A, Dipasquale M. Assessment and treatment of the vegetative and minimally conscious patient. In: Rosenthal M, Kreutzer JS, Griffith ER, et

al., editors. Rehabilitation of the adult and child with traumatic brain injury. Philadelphia, PA; 1999. p. 435–52.

[19] Giacino JT, Kalmar K, Whyte J. The JFK Coma Recovery Scale-Revised: measurement characteristics and diagnostic utility. Arch Phys Med Rehabil 2004;85:2020–9. http://dx.doi.org/10.1016/j.apmr.2004.02.033.

[20] Wannez S, Heine L, Thonnard M, Gosseries O, Laureys S, Coma Science Group collaborators. The repetition of behavioral assessments in diagnosis of disorders of consciousness: repeated CRS-R assessments for diagnosis in DOC. Ann Neurol 2017;81:883–9. http://dx.doi.org/10.1002/ana.24962.

[21] Luaute J, Maucort-Boulch D, Tell L, Quelard F, Sarraf T, Iwaz J, et al. Long-term outcomes of chronic minimally conscious and vegetative states. Neurology 2010;75:246–52. http://dx.doi.org/10.1212/WNL.0b013e3181e8e8df.

[22] Thibaut A, Bruno M-A, Ledoux D, Demertzi A, Laureys S. tDCS in patients with disorders of consciousness: sham-controlled randomized double-blind study. Neurology 2014;82:1112–8. http://dx.doi.org/10.1212/WNL.0000000000000260.

[23] Demertzi A, Ledoux D, Bruno M-A, Vanhaudenhuyse A, Gosseries O, Soddu A, et al. Attitudes towards end-of-life issues in disorders of consciousness: a European survey. J Neurol 2011;258:1058–65. http://dx.doi.org/10.1007/s00415-010-5882-z.

[24] von Elm E, Altman DG, Egger M, Pocock SJ, Gøtzsche PC, Vandenbroucke JP. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. Epidemiology 2007;18:800–4. http://dx.doi.org/10.1097/EDE.0b013e3181577654.

[25] Giacino JT, Katz DI, Schiff ND, Whyte J, Ashman EJ, Ashwal S, et al. Practice guideline update recommendations summary: disorders of consciousness. Neurology 2018;91:450–60. http://dx.doi.org/10.1212/WNL.0000000000005926.

[26] Giacino JT, Katz DI, Schiff ND, Whyte J, Ashman EJ, Ashwal S, et al. Practice guideline update recommendations summary: disorders of consciousness. Arch Phys Med Rehabil 2018;99:1699–709. http://dx.doi.org/10.1016/j.apmr.2018.07.001.

[27] Naccache L. Minimally conscious state or cortically mediated state? Brain 2018;141:949–60. http://dx.doi.org/10.1093/brain/awx324.

[28] Stender J, Gosseries O, Bruno M-A, Charland-Verville V, Vanhaudenhuyse A, Demertzi A, et al. Diagnostic precision of PET imaging and functional MRI in disorders of consciousness: a clinical validation study. The Lancet 2014;384:514–22. http://dx.doi.org/10.1016/S0140-6736(14)60042-8.

[29] Bodart O, Gosseries O, Wannez S, Thibaut A, Annen J, Boly M, et al. Measures of metabolism and complexity in the brain of patients with disorders of consciousness. NeuroImage Clin 2017;14:354–62. http://dx.doi.org/10.1016/j.nicl.2017.02.002.

[30] Kunieda K, Ohno T, Fujishima I, Hojo K, Morita T. Reliability and validity of a tool to measure the severity of dysphagia: the food intake LEVEL Scale. J Pain Symptom Manage 2013;46:201–6. http://dx.doi.org/10.1016/j.jpainsymman.2012.07.020.

[31] Shaker R, editor. Principles of deglutition: a multidisciplinary text for swallowing and its disorders.. New York: Springer; 2013.

[32] Sessle BJ, Adachi K, Avivi-Arber L, Lee J, Nishiura H, Yao D, et al. Neuroplasticity of face primary motor cortex control of orofacial movements. Arch Oral Biol 2007;52:334–7. http://dx.doi.org/10.1016/j.archoralbio.2006.11.002.

[33] Sessle BJ, Yao D, Nishiura H, Yoshino K, Lee J-C, Martin RE, et al. Properties and plasticity of the primate somatosensory and motor cortex related to orofacial sensorimotor function. Clin Exp Pharmacol Physiol 2005;32:109–14. http://dx.doi.org/10.1111/j.1440-1681.2005.04137.x.

[34] Avivi-Arber L, Martin R, Lee J-C, Sessle BJ. Face sensorimotor cortex and its neuroplasticity related to orofacial sensorimotor functions. Arch Oral Biol 2011;56:1440–65. http://dx.doi.org/10.1016/j.archoralbio.2011.04.005.

[35] Dellow PG, Lund JP. Evidence for central timing of rhythmical mastication. J Physiol 1971;215:1–13.

[36] Yamada Y, Yamamura K, Inoue M. Coordination of cranial motoneurons during mastication. Respir Physiol Neurobiol 2005;147:177–89. http://dx.doi.org/10.1016/j.resp.2005.02.017.

[37] Moore JD, Kleinfeld D, Wang F. How the brainstem controls orofacial behaviors comprised of rhythmic actions. Trends Neurosci 2014;37:370–80. http://dx.doi.org/10.1016/j.tins.2014.05.001.

[38] Mazzone SB, Cole LJ, Ando A, Egan GF, Farrell MJ. Investigation of the neural control of cough and cough suppression in humans using functional brain imaging. J Neurosci 2011;31:2948–58.

[39] Hamdy S, Aziz Q, Thompson DG, Rothwell JC. Physiology and pathophysiology of the swallowing area of human motor cortex. Neural Plast 2001;8:91–7. http://dx.doi.org/10.1155/NP.2001.91.

[40] Martin RE, Sessle BJ. The role of the cerebral cortex in swallowing. Dysphagia 1993;8:195–202. http://dx.doi.org/10.1007/BF01354538.

[41] Matthews CT, Coyle JL. Reducing pneumonia risk factors in patients with dysphagia who have a tracheotomy: what role can SLPs play? ASHA Lead 2010;15, https://leader.pubs.asha.org/doi/full/10.1044/leader.FTR4.15062010.np.

[42] Roberts H, Greenwood N. Speech and language therapy best practice for patients in prolonged disorders of consciousness: a modified Delphi study. Int J Lang Commun Disord 2019;54:841–54. http://dx.doi.org/10.1111/1460-6984.12489.

[43] Miles A, Hunting A, McFarlane M, Caddy D, Scott S. Predictive value of the New Zealand Secretion Scale (NZSS) for pneumonia. Dysphagia 2018;33:115–22. http://dx.doi.org/10.1007/s00455-017-9841-z.