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A multiple-case study testing the implementation of a non-aphasia-specific app into evidence-based therapy

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Digital treatments on tablet computers have become increasingly popular to deliver speech and language therapy. Practice guidelines have been proposed to successfully integrate non-aphasia-specific apps into rehabilitation, but few evidence-based reports are available yet. Three individuals with acquired language disorders trained at home with a mainstream app containing personalized material. The treatment plan was specific to each individual and supervised by a speech and language therapist. All three participants showed significant improvements in picture naming that were specific to the treated items and treatment gains were overall maintained after a couple of months. Treatments carefully designed and delivered in an app led to specific language improvements similar to those previously reported in the literature with or without technology. There is presently no proof that ready-to-go dedicated apps are more effective than this kind of mainstream app allowing the creation and adaptation of materials and tasks to evidence-based knowledge.

Keywords: aphasia, anomia, rehabilitation, technology, iPad

De plus en plus de thérapies orthophoniques sont proposées sur tablette tactile. Des recommandations ont été publiées pour intégrer des applications non spécifiques à l'aphasie dans la réhabilitation, mais il existe encore peu d'études concluantes à ce sujet. Trois personnes présentant des troubles acquis du langage se sont entrainées à domicile avec une application contenant du matériel personnalisé. Un plan de traitement spécifique élaboré pour chaque participant a été supervisé par un orthophoniste. Les trois participants ont montré des améliorations significatives en dénomination d'images, spécifiques aux items travaillés et stables après quelques mois. Des traitements élaborés soigneusement et administrés à l'aide d'une application ont engendré des progrès similaires à ceux rapportés dans la littérature avec ou sans technologie. Il n'y a actuellement aucune preuve que des applications prêtes à l'emploi soient plus efficaces que des applications permettant la création et l'adaptation du matériel et des tâches aux connaissances basées sur les preuves.

Mots-clés : aphasie, anomie, réhabilitation, technologie, iPad

Introduction

Aphasia is commonly observed after brain damage and has a substantial impact on quality of life (Lam & Wodchis, 2010). Therefore, recovery of language functions is a major challenge for people with aphasia and their entourage. In addition to traditional speech and language therapy, aphasic persons increasingly benefit from digital therapies in clinical settings or at home. Indeed, the use of technology is an amazing way of reaching adequate treatment intensity, known as a key factor of aphasia therapy effectiveness (Brady et al., 2016). Since the first group studies on computerized speech therapy published nearly thirty years ago (e.g., Stachowiak, 1994), technological devices have become incredibly popular, more affordable and are an integral part of our daily lives. Indeed, most individuals of all ages living in developed countries now have access to high technology devices such as computers, tablets, or smartphones at home. Thanks to dedicated software directories/stores in tablets and smartphones, disseminating and installing new software or apps has never been as accessible as it is today. Moreover, combining standard treatments with technology-based self-therapy could reduce health-care costs by about half according to the estimates of a recent study conducted in the UK (Palmer et al., 2019).

In the management of aphasia, many technologyenhanced treatments have proved their effectiveness to treat anomia (Lavoie et al., 2017), but also reading and comprehension deficits (cf. Zheng et al., 2015). Besides restoring language skills, technologies can also for instance in allowing augmentative and alternative communication (Taylor et al., 2019) or by using voice recognition and word prediction to support writing (e.g., Dietz et al., 2011; Marshall et al., 2019). According to Macoir et al. (2019), the effectiveness of self-administered

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technology-based treatments depends on three factors related to 1) the treatment content; 2) the technology; and 3) the patient. Concerning treatment content, it should be implemented by a clinician to target specific objectives and ideally the software should adjust the complexity of the task to the actual performance and give appropriate feedback. In terms of technology, the software must be aphasia-friendly (i.e., adapted to language/cognitive deficits) and familiarization sessions should be provided. Finally, concerning the user, he/she must be able and motivated to use technology, and so must their environment. Contrary to presumed barriers, it seems that usage of digital speech and language therapies is limited neither by age nor by geographical remoteness (Munsell et al., 2020).

Among all technological devices on the market, tablet computers such as iPads are increasingly used in neurorehabilitation and more especially in speech and language therapy (Ameer & Ali, 2017). Some therapeutic apps are available nowadays in mainstream popular tablet app-stores and are able to improve language skills of aphasic persons (e.g., Constant Therapy app, Des Roches et al., 2015 (https://thelearningcorp.com/constant-therapy/); Language Therapy app, Stark & Warburton, 2018 (https://tactustherapy.com/app/language/); Steele et al., 2015; TalkPath Therapy app, cf. Repetto et al., 2020 (https://therapy.aphasia.com)). However, these ready-to-use apps are rarely customizable (e.g., training items are already chosen and might be useless for the person), they do not provide relevant settings to match the material to the underlying deficit (e.g., targeting only words of a given length) and they often are only reliably available in English. In addition, they are rather expensive thus creating inequalities in access to care. It should also be noted that several aphasia specific treatment apps are still in the research stage and not available to the community yet (e.g., Gerber et al., 2019). Personalization to individual needs and adjustment of the level of difficulty are crucial just to adhere to the therapy, as ready-to-use non-customizable apps are inevitably judged inappropriate (i.e., too easy or too difficult) by the users (Pugliese et al., 2019). As a matter of fact, it is also possible to incorporate non-aphasia-specific apps into the therapy, but the intended software must pass three filters according to Ramsberger and Messamer (2014): 1) the language filter (i.e., the tasks, the approach, and the focus must be appropriate and adapted to the language profile); 2) the non-linguistic capabilities filter (i.e., the app must be compatible with sensory, motor, and cognitive abilities); and 3) the technology filter (i.e., the user has to possess a compatible device and internet connection if applicable). In their best practice recommendations, Ramsberger and Messamer (2014) outline several non-aphasia-specific apps that can be implemented in therapies and propose to use the app Bitsboard (https://bitsboard.com) to assess how aphasic people can handle an iPad. However, this app could also serve as a therapeutic tool, as it has already proved successful to improve word comprehension in French in children with developmental language disorders (cf. Durrleman et al., 2019) and possibly speech production in Chinese (cf. Hsueh-Min & Yu-Chih, 2017).

Bitsboard is a mainstream multilingual free iPad app which is not particularly designed for aphasia. According to its website, Bitsboard is an app “to learn, to teach and to play”: more than forty games are available and particular attention has been given to accessibility/settings for users with special needs. The main strength of the app is its high level of customization: training items can rapidly be constructed by choosing personal pictures (i.e., any self-made picture on the iPad or available on the internet) and linking them with a written word/sentence, a written description and self-recorded audio in any language. Items are grouped in folders named “boards”, that serve as stimuli packages for all games. Boards can be shared between users in a convenient online catalog or by email. Importantly, every game comes with plenty of parameters (e.g., number of items/repetitions, item randomization, amount and type of cues) and flexible options to automatically adjust the level of difficulty (e.g., begin with two distractors pictures and add up to six foils in the absence of errors). At the end of each game, a colorful feedback listing the failed/successful items appears alongside a success rate that is kept in the Statistics section of the app to track the progress. Currently, applications such as Bitsboard may be appropriate for a large panel of aphasic persons, given the extensive settings available to customize the app and to easily create tailored material to meet individual needs.

The aim of the present exploratory study is to test whether the app Bitsboard can be used to implement evidence-based therapies in clinical practice with French-speaking individuals. As game-based interventions are quite motivating for aphasic speakers without compromising treatment outcomes (Romani et al., 2018), it is hypothesized that Bitsboard will lead to significant improvement of language functions.

Methods

Participants

Three adults with acquired language impairments were included in the present investigation: they were attending traditional speech therapy sessions and they agreed to use the free Bitsboard app on their own iPad for self-administered training at home.

P1 is a 56-year-old man presenting with transcortical sensory aphasia following a left hemispheric stroke in middle and anterior arterial territories. One year and a half after stroke, his anomia was still moderate to severe and most likely due to the deregulation of semantic cognition. His oral speech production was fluent, but not always informative due to semantic, formal, and unrelated paraphasias, as well as dyssyntactic alterations. He also presented with deep agraphia and deep alexia. P1 benefited from intensive speech and language therapy (i.e., 5-10 hours per week) as an inpatient during the first 5 months post-stroke. Then as an outpatient, he benefited from speech and language therapy for 2 hours per week up to 1.5 years post-stroke.

P2 is a 50-year-old woman presenting with severe Broca aphasia. Two years after her left hemispheric ischemic stroke in the middle arterial territory, her writing deficit was still severe due to surface agraphia and anomia. Her oral speech production was agrammatic, with phonemic and semantic paraphasias. She also suffered from deep alexia. P2 benefited from intensive speech and language therapy (i.e., 5-8 hours per week) as an inpatient during the first 3 months post-stroke. Then as an outpatient, she benefited from speech and language therapy for 4-5 hours per week for 5 months and 2-3 hours per week up to two years post-stroke.

P3 is a 75-year-old man presenting with mild cognitive impairment due to a left fronto-temporal neurodegenerative disease under investigation. His main subjective concern was about retrieving the names and surnames of his relatives. His oral speech production was fluent and informative without anomia on verbs or common nouns. Writing and reading skills were preserved. No speech and language therapy were administered before the treatment reported here.

Materials

The material was constructed on the paid version Bitsboard Pro by speech and language therapists and then transferred to the iPad of the participant on which the free version of Bitsboard was installed. The stimuli were personalized for each participant:

For P1, 144 color photographs corresponding to common nouns from 8 semantic categories were selected in the Bank of Standardized Stimuli (Brodeur et al., 2010) and Google Images. They were divided into two lists of 72 items matched in terms of word frequency (according to New, B. & Pallier, C. (2021). Lexique. Retrieved from: www.lexique.org) and length (i.e., number of phonemes per word). Each picture was linked to a strongly associative word (e.g., plate – food), according to an online questionnaire filled in by 20 healthy controls;

For P2, 90 color photographs corresponding to common nouns were selected on Google Images and divided into two lists of 45 items matched in terms of word frequency, number of letters per item and regularity;

For P3, 32 color photographs of far relatives (list A, *n* = 16) and close relatives (list B, *n* = 16) were brought by the participant.

Treatment design

The treatment target and the tasks/games were adapted to the needs and possibilities of each participant.

For P1, the commonly defined objective was to improve the retrieval of common nouns by means of picture naming and to test whether a semantic association task in addition to picture naming could boost the gains of the treatment. Each list of 72 items was assigned to a different therapy condition (A and B below) and trained in a sequential crossover design after a multiple baseline:

The treatment tasks for list A (*n* = 72) were lexical-semantic association followed by picture naming, by means of Bitsboard games Pop Quiz (picture-word association), Review (oral picture naming), and Spelling Bee (written picture naming). P1 always began with the picture-word association task, in which he saw a picture with two written words underneath. He had to choose the word (e.g., food) semantically associated with the picture (e.g., plate) and the distractor was randomly selected by Bitsboard. Then P1 could choose to continue with oral or written picture naming. In the oral picture naming task, P1 had to name aloud the picture presented on the screen and press the screen at his own pace to hear the correct answer, before self-judging how he performed the trial (he had to choose between three color buttons: red = badly, yellow = incompletely, green = well). In the written picture naming task, P1 had to write the name of the picture presented on the screen and he could briefly see the written answer for 2 seconds by pressing a help button at any time and as often as necessary (delayed copy), with instant visual and auditory feedback if he pressed a wrong letter.

The treatment task for list B (*n* = 72) was picture naming only, with Review (oral picture naming) and Spelling Bee (written picture naming) Bitsboard games.

After two pre-tests evaluating oral picture naming of the 144 items separated by a 3-week interval, P1 trained at home first with therapy A (lexical-semantic associations + picture naming) for 18 sessions over 3 weeks, then a post-test with the 144 items to name was administered and he trained at home with therapy B (picture naming only) for 18 sessions over 3 weeks. Right after the end of therapy B, a post-test was again administered on the 144 items, with follow-ups conducted 3 months and 1 year after the end of both therapies. During testing phases, responses were rated as correct if the target word was given within 10 seconds and self-corrections were accepted.

For P2, the commonly defined objective was to improve the retrieval of orthographic forms of common nouns. A list of 45 items was trained in a multiple baseline design with the other (untrained) list serving as control. Four Bitsboard games were practiced in random order:

Spelling Bee: written picture naming task, where the name of the picture presented on the screen has to be written, with the possibility of pressing a help button to briefly see the written answer for 2 seconds at any time and as often as necessary (delayed copy), with instant visual and auditory feedback when pressing a wrong letter.

Word Builder: anagrams of the words, where the letters must be put in the right order (n.b., distractor letters were disabled here but can also be added in the settings).

Missing Letter: written words presented with a letter missing in a random position (n.b., the game can also focus only on first, middle, or last letters depending on the settings).

Word Search: three words hidden in a grid that need to be highlighted (n.b., the size of the grid, number of words, reading direction and visual help in finding the first letter can be adapted in the settings).

After two pre-treatment assessments evaluating written picture naming of the 90 items separated by one month, P2 trained at home with list A for 20 sessions over 2.5 months. Right after the end of list A training, a post-test was administered on the 90 items with a follow-up 4 months later. During testing phases, responses were rated as correct if the target word was written correctly (even if self-corrected), without time limit.

For P3, the commonly defined objective was to improve the retrieval of proper nouns and more particularly names and surnames of relatives. The two lists were trained in a multiple baseline crossover design by means of two Bitsboard games. Within each session, P3 trained first with the Pop Quiz game consisting in matching a picture of a person's face with his/her name and surname among distractors randomly chosen by the app. Face-to-name matching began with a forced choice between 2 surnames/names and automatically adapted to the performance of P3: after 2 consecutive correct trials, another written distractor was introduced to a maximum of 6 surnames/names presented under the picture, but in case of a mistake, one distractor was removed in the next trial. Second, P3 trained with Spelling Bee, in which he had to write the surname and the name of the depicted familiar face, with the possibility to press a help button to briefly see the written answer for 2 seconds and as often as necessary (delayed copy), with instant visual and auditory feedback when pressing a wrong letter.

After a single baseline evaluating the surname/name retrieval of the 32 people, P3 trained at home first with list A (16 far relatives) for 10 sessions over 1 month, then a post-test with the 32 faces to name was administered and he continued with list B (16 close relatives) for another 10 sessions over 1 month. Right after the end of list B training, a picture naming post-test was again administered on the 32 faces. The follow up 4 months after the therapy was conducted on the same set of 32 faces. During testing phases, P3 became 2 points if he retrieved both the surname and the name of the person and 1 point if he retrieved only one of these. Self-corrections were accepted without time limit.

Common procedure and statistical analyses

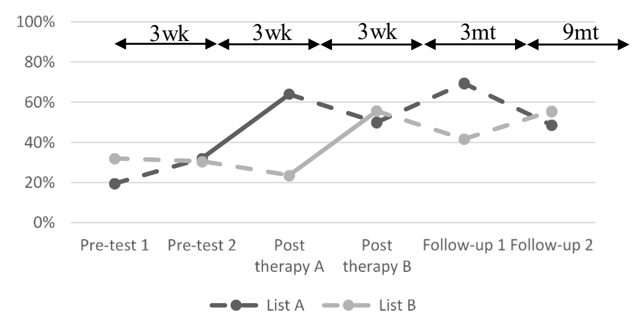
The first treatment session was entirely conducted with the participants in order to show them every exercise with multiple examples. A printed paper with very simple written procedures and corresponding iPad/Bitsboard icons was given to them after this first session. Therapy was then self-administered at home, with regular monitoring in face-to-face setting to ensure that the tasks were performed correctly and to verify how many sessions were launched at home, as well as the reached accuracy thanks to the results recorded in Bitsboard. For statistical analysis, Friedman chi square tests (for more than two time-points) and Wilcoxon signed rank tests (for two time-points) were used to evaluate the impact of the intervention. Because multiple comparisons were made on the same dataset (by pairs between the time-points), results were considered significant with an alpha criterion below $p = .01$ (i.e., $p = .05$ divided by 5) for P1 and below, $p = .017$ (i.e., $p = .05$ divided by 3) for P2 and P3, according to the conservative Bonferroni correction for family wise errors.

Results

For P1 (cf. Figure 1), Friedman chi square test on six time-points revealed significant changes across time for the first list / therapy A (c2 = 28.88, $p < .001$) and for the second list / therapy B (c2 = 17.82, $p = .001$). Wilcoxon signed rank tests were computed between each pair of consecutive time-points. For the first list of 72 items (therapy A), significant positive differences were found pre- and post-treatment ($p < .001$) and between the first and the second follow-up ($p = .003$), whereas a negative difference appeared between the second and the third follow-up ($p = .007$). For the second list of 72 items (therapy B), the only difference that resisted the threshold was the treatment phase ($p < .001$). In sum, P1 showed significant improvements in picture naming, that were specific to the trained material and to the treatment phase and maintained up to one year after the end of the treatment for the second list/ therapy B. Improvements for the first list/therapy A showed some variations over time.

Figure 1

P1's naming accuracy across time-points for each sequentially treated list



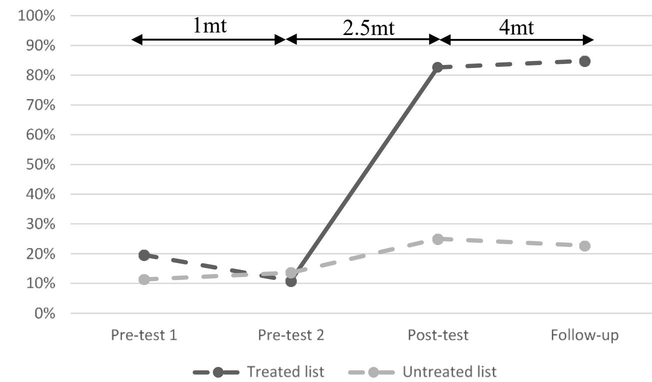
Note. Treatment phases are represented with solid lines and treatment-free periods with dashed lines; time intervals are given in weeks (wk) or months (mt).

For P2 (cf. Figure 2), Friedman chi square test revealed significant changes across time for the treated list (c2 = 80.59, $p < .001$) but not for the untreated list (c2 = 4.13, $p = .13$). Wilcoxon signed rank tests were computed between each pair of consecutive time-points for the treated condition. Changes were significant between the second pre-test and the post-test ($p < .001$), but not between the two pre-tests ($p = .23$) and neither between the post-test and the follow -up ($p = .81$). In sum, P2 showed significant improvements in picture naming, that were specific to the treated list without generalization to untreated items. Gains for the treated list were maintained 4 months after the end of therapy.

For P3 (cf. Figure 3), Friedman chi square test revealed significant changes across time for the first list (c2 = 20.18, $p < .001$) and for the second list (c2 = 14.14, $p = .003$). Wilcoxon signed rank tests were computed between each pair of consecutive time-points for the treated condition. For both lists, the only difference that resisted the threshold was the treatment phase ($p = .007$ for the first list and $p = .01$ for the second list). In sum, P3 showed significant improvements in naming persons, that were specific to the treatment phases. No generalization was found to

Figure 2

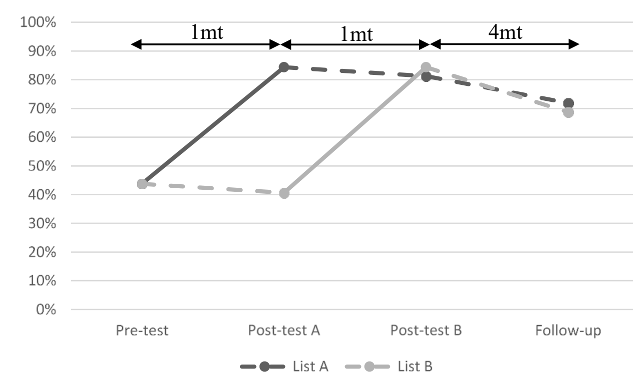
P2's naming accuracy across time-points for the treated and the untreated lists



Note. Treatment phases are represented with solid lines and treatment-free periods with dashed lines.

Figure 3

P3's naming accuracy across time-points for each sequentially treated list



Note. Treatment phases are represented with solid lines and treatment-free periods with dashed lines.

untreated items and gains were maintained 4 months after the end of the treatment.

Discussion

The aim of the present investigation was to assess if a non-aphasia-specific app was appropriate to implement evidence-based treatments in clinical practice. As P1, P2, and P3 significantly improved in naming treated items in the medium/long term, it seems that Bitsboard was an efficient tool to support self-administered speech and language therapy at home. Indeed, the three criteria predicting the success of self-administered digital treatments by Macoir et al. (2019) were met here: the treatment content was implemented by a clinician, specific and individual objectives were targeted and a certain amount of help, feedback and adjustment of difficulty level were provided. Importantly, the minimalistic user interface of Bitsboard is aphasia-friendly if the clinician hides unnecessary features and provides training sessions. All three participants were motivated and able to invest time and energy in their digital treatment. This was even the case for P2, who was unfamiliar with tablet computers before the treatment.

For P1, gains were stable after therapy B (picture naming alone) but volatile after therapy A (which combined semantic associations and picture naming): even if immediate treatment effects were significant, accuracy decreased at the first follow-up three months post-therapy and increased again one year post-therapy. Such fluctuations could be explained by the refractory state hypothesis accounting for inconsistent performance across testing sessions in individuals with "semantic aphasia" (Warrington & Mccarthy, 1983). In a more recent variant of the refractoriness hypothesis, it has been demonstrated that persons with "semantic aphasia" typically produce associative paraphasias (e.g., "hump" for "camel"), because they are struggling to inhibit strong (yet task-irrelevant) associations due to a loss of semantic control (Jefferies & Lambon Ralph, 2006). Indeed, P1 produced numerous associative paraphasias throughout the testing sessions. It was thus probably not a good option to train the matching of pictures with strong associates (e.g., bird-nest) instead of classical picture-word matching for P1. Even if some previous treatment studies using Semantic Feature Analysis (Boyle & Coelho, 1995) argued that providing semantic associations was ineffective for patients with semantic deficits (e.g., van Hees et al., 2013), another more recent randomized controlled trial with larger groups of patients was less conclusive (Kendall et al., 2019). For P1 and potentially for other individuals with semantic deregulation/control loss, picture naming seems to provide more stable improvements when practiced without adding lexical-semantic associations. Crucially, comparing these two lists/therapies with Bitsboard shed light on which type of treatment should be preferred in the future for P1.

For P2, the results showed significant and stable gains after training with four different exercises targeting the retrieval of orthographic forms in various ways. These gains were likely due to strengthening either the mapping between semantic and orthographic representations (Spelling Bee game) or the orthographic representations themselves (Word Builder, Missing Letter, and Word Search games). As P2 trained randomly with all four exercises, it is unfortunately impossible to identify which/if a particular game improved written word retrieval the most. It would be interesting for future research to tease apart the contribution of every single game from the combination of the four games.

For P3, significant and stable improvements occurred in proper noun retrieval, despite degenerative brain damage. These improvements were probably due to reinforcement of the links between semantic representations (faces) and phonological/orthographic representations (names), with word-to-picture matching (Pop Quiz game) facilitating picture naming of the same items thereafter (Spelling Bee game).

To exemplify alternative potential uses of Bitsboard that we tested so far with other brain injured adults in clinical practice, we could successfully restore the phonological-to-graphemic conversion by means of linking each grapheme with a key word, improve word discrimination of minimal pairs, ameliorate sentence production of short utterances, and reduce verb anomia. These other examples in which we used Bitsboard are not detailed here, because the app was used in complement to parallel standard speech therapy sessions and exercises, therefore rendering impossible any conclusion about the efficacy of Bitsboard only. In addition to that, it is worth noting that Trace It game may be of great help for apraxic dysgraphia (i.e., use of a stylus to draw the letters with decreasing cues), Listen Up game for verbal short-term memory training (i.e., word span of increasing length), Say It game for picture naming or speech motor disorders with objective feedback (i.e., speech recognition feature) and Questions game to create any new exercise.

Bitsboard passes through the linguistic and non-linguistic filters mentioned by Ramsberger and Messamer (2014). Concerning the language filter, the tasks and the focus can/must be selected by the clinician and adjusted to the underlying language impairment. Regarding the non-linguistic filter, Bitsboard provides many accessibility settings to make it compatible to various brain-damaged profiles (e.g., non-target items and games can be hidden, feedbacks can be set to immediate vs. delayed, next trials can be launched automatically, swipe/tap/drag movements can alternately be selected as responses, etc.). However, the third filter about technology is the most delicate, as Bitsboard is currently only available for iOS devices (iPhones and iPads), but on the positive side it does not require an internet connection after installation – and it is free.

The present case reports suggest that it is feasible to use an app such as Bitsboard in an efficient way with brain-damaged individuals suffering from aphasia due to various etiologies (focal stroke or neurodegenerative atrophy). One limitation that deserves future attention is the lack of evaluation about the transfer to ecological situations, as a large study recently indicated that transfer to conversation settings was not straightforward after anomia therapy (Palmer et al., 2019). Another limitation of the present report concerns the sample size, like most case studies using digital therapies: large randomized control trials are welcome and could incorporate neuroimaging tools to highlight treatment-related effects at the brain level (Choi et al., 2019) or the added value of neurostimulation techniques.

There is currently no proof that dedicated aphasia apps are more effective than mainstream apps allowing the creation of materials and task adaptations to evidence-based knowledge supervised by a clinician. Bitsboard is probably not a unique case and several other non-aphasia specific apps could be of great help in language rehabilitation, even though finding appropriate software among the millions of apps available is challenging and time-consuming. The key component is to adopt the same evidence□based reasoning than in any standard treatment implementation. Actually, most of 1.0 paper -based therapies that have proven to be effective could be adapted to 2.0 technology-based settings such as Bitsboard.

Conclusion

A free mainstream iPad app led to specific language improvements similar to those previously reported in the anomia literature with or without technology. This highly customizable app holds the potential to implement aphasia treatment in several languages. Technology is an ideal support to enhance the intensity of speech therapy and the present case reports confirm that non-aphasia apps responding to some criteria (Macoir et al., 2019) and to some filters (Ramsberger & Messamer, 2014) can support evidence-based treatments. In the future, the addition of self-administered digital therapies alongside face-to -face sessions should become the standards and will hopefully be soon referred to as “traditional” speech therapy

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