A Novel Bone Conduction Hearing System May Improve Memory Function in Children with Single Side Hearing loss: A Case-Control Study

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OBJECTIVES: To evaluate the effects of an adhesive adapter prosthesis (AAP) on memory function in pediatric subjects with single side hearing loss (SSHL).

MATERIALS and METHODS: Case-control study. 19 pediatric subjects with mild to moderate SSHL treated with AAP and 15 subjects with normal hearing (control group) were included in this study. Working and short-term memory functions were tested in all subjects, in silence and noise conditions. In SSHL subjects, tests were performed before the AAP was applied (T0) and at 1-month (T1) follow-up. The control group was tested once.

RESULTS: AAP significantly improved working memory function in noise as measured at T1 (p<0.01) compared with T0, but T1 scores in children with SSHL remained significantly different from the ones of the control group (p<0.01). AAP also significantly improved short-term memory function test scores at T1 compared with T0 (p<0.01), but despite being in the normal range for the subjects' age, the scores remained significantly different from those of the control group (p<0.01).

CONCLUSION: In pediatric subjects with mild, moderate, and moderate-severe SSHL, restoration of bilateral hearing through AAP improved short-term memory function and working memory function in noise, as measured at 1 month follow-up; however, AAP did not seem to lead to a full restoration of such functions as measured by a comparison with healthy controls. Further studies with longer follow-ups might help elucidate whether AAP can elicit further improvements in memory functions.

KEYWORDS: Conductive single side hearing loss, bone conduction hearing aid, working memory, short-term memory

INTRODUCTION
The importance of recovering bilateral hearing in patients with bilateral deafness is widely agreed upon. Conversely, subjects with single side hearing loss (SSHL) are rarely treated with hearing aids (1), bone anchored hearing aids (2), or cochlear implants (CIs) (3). In fact, in SSHL patients, a good unilateral hearing function is typically considered acceptable, and the option of using a hearing aid is often underexplored. However, bilateral hearing function is not only important for hearing correctly but it is also necessary for identifying the direction of sound (4), perceiving nuances of music (5), and improving hearing ability in noisy situations (2,3); moreover, in children, it helps the development of a normal auditory pathway (6). As recently shown, improvement in bilateral hearing translates into improvement in quality of life (7), social life (7,8), speech perception (8), and memory function (9).

Several studies have shown that in pediatric subjects, hearing restoration improves memory function because of the key role of hearing in brain development (10-13). In 2018, Di Stadio et al. (10) investigated the effects of a bone anchored hearing implant on speech perception, dictation, and memory in children with single side deafness (SSD) (10) and showed that the implant ul-
timely improved the subjects’ memory performance, confirming that restoration of bilateral hearing function stimulates memory function improvement/development \[20\]. However, although the link between hearing function and memory function has been demonstrated \[9, 10-15\] and bilateral hearing implantation has been shown to improve the quality of life in pediatric subjects \[3, 4-8\], as of today, mild and severe SSHL are rarely treated with hearing aids in children.

Adhesively attached prosthesis (AAP) (ADHEAR\textsuperscript®, Medel International, Milano, Italy, www.medel.com) is a new generation bone hearing aid system that does not require surgery and is attached to the patient’s temporal bone, superiorly to the pinna, with an adhesive patch (Figure 1). Its audiological and clinical indications are similar to those of other bone anchored commercial prostheses or head bands. It can be used for treating subjects with SSHL, either acquired or congenital, with bone threshold equal to or smaller than 25 dB. AAP uses 2 microphones for directionality and a digital signal processor. As shown by Dobrev et al. \[16\], the particular location at which it attaches to the patient’s mastoid allows a constant and stable stimulation of the cochlea, ultimately leading to higher performance than traditional bone anchored hearing prostheses. The effects of AAP have overall been little investigated \[15\]. Several prospective, randomized studies have shown that AAP improves speech understanding in noise and sound localization \[17, 18\] but whether and how AAP ultimately affects memory function in children with various degrees of severity of SSHL is still unclear. Thus, this study aimed at filling this gap in the literature and at evaluating the effects of AAP on memory function in pediatric subjects with various degrees of severity of SSHL.

**MATERIALS AND METHODS**

This case-control study was conducted in the Cochlear Implantation Center of the Santobono-Pausilipon Children’s Hospital, Naples, Italy, from June to December 2017. All study procedures were approved by the Institutional Review Board (IRB) of the hospital.

**MAIN POINTS**

- The use of a prosthesis to restore bilateral hearing function in patients with single side hearing loss (SSHL) should always be considered.
- Bilateral hearing restoration positively impact working memory function especially in noise condition.
- Improvement of memory function positively impacts brain development; thus, it is extremely important in children.
- The use of an adhesive anchored prosthesis allows recovery of working memory function in patients with single side hearing loss, although recovery might not be as good as that obtained with a bone anchored hearing implant (BAHI).
- For SSHL patients that are candidates for surgical implantation of BAHI or cochlear implant an adhesive anchored prosthesis might be a useful tool to test the potential efficacy of such hearing aids prior to surgery.

19 children (12 male and 7 female, average age 6.3 years [SD: 1.24; CI 95%: 5-8]) with mild (26-40 dB), moderate (41-55 dB), and moderate-severe (56-70) SSHL and normal hearing threshold in the contralateral ear as measured by Pure Tone Audiometry (PTA) testing were enrolled in the study (Table 1). In addition, 15 children (9 male and 6 female, average age 6.5 years [SD: 1.18; CI 95%: 5-8]) with normal bilateral hearing thresholds were enrolled as control group (CG). All children were native Italian speakers.

Memory performances were evaluated before (T0) and 4 weeks after (T1) the attachment of ADHEAR\textsuperscript® in the subjects with SSHL (none of the subjects received speech rehabilitation) and only once in the subjects of the CG. Both working and short-term memory functions were evaluated, as detailed below. All tests were conducted by a speech therapist and a psychologist, both with more than 10 years of experience.

**Working Memory Testing**

Working memory (WM) is the part of memory that manages received information. It allows us to understand the meaning of a whole sentence even when we don’t understand each single word \[19\].

Working memory evaluation was performed with PROMEA battery of tests. Subjects were asked to repeat “non-words” sentences and count backward from 10 to 1 \[20\]. A “non-words” sentence is a sentence composed of common words and other words that have a sound similar to that of existing words in the (Italian) language but in fact do not mean anything (for example “sasta” has a sound similar to “pasta” \[20\] but does not mean anything). A subject with a properly functioning WM will correct the “non-word” “sasta” and repeat “pasta,” indicating that she/he understands the meaning of the whole sentence.

All tests were performed in quiet and noise (cocktail party noise) \[21\].

The tests scores were calculated as the number of correct answers divided by the total number of questions (percentage). Following Bisiacchi, the total number of questions was set to 39 \[22\]; also, 0-25%, 26-50%, 51-75%, and 76-100% of correct answers indicated a severe deficit, moderate deficit, sufficient, and excellent WM function, respectively \[22\]. Note that the Bisiacchi version of the test was chosen because it specifically measures memory function in children of age between 5 and 8 years \[22\].

**Short-Term Memory Testing**

The short-term memory is the part of the memory that stores information for a short time, typically 10-15 seconds \[23\].

Evaluation of short-term memory function was performed in a manner similar to the evaluation of WM function (see above), but the subject was asked to repeat the last 3 words of a sentence exactly how she/he heard them. For example, the sequence of words “la sasta al pomodoro” (“pasta with tomato sauce”) had to be repeated as it was, without correcting the wrong word (i.e., “sasta”). This test was performed in quiet only, as noise may affect short-term memory function and the subject’s ability to hear a “non-word.”
Table 1. Patients’ demographics and PTA data

| Case | Sex | Age (y) | Side | Type of HL | Etiology     | PTA BC (dB) |
|------|-----|---------|------|------------|--------------|-------------|
| 1    | M   | 8       | R    | SNHL       | Idiopathic   | 70          |
| 2    | M   | 8       | R    | CHL        | Post-tympanoplasty | 35          |
| 3    | F   | 7       | L    | SNHL       | Idiopathic   | 45          |
| 4    | M   | 8       | R    | SNHL       | Congenital   | 55          |
| 5    | M   | 5       | R    | SNHL       | Idiopathic   | 65          |
| 6    | F   | 5       | R    | SNHL       | Congenital   | 50          |
| 7    | F   | 6       | R    | CHL        | Post-tympanoplasty | 40          |
| 8    | F   | 6       | R    | SNHL       | Congenital   | 60          |
| 9    | M   | 4       | R    | CHL        | Congenital   | 40          |
| 10   | M   | 6       | R    | SNHL       | Idiopathic   | 50          |
| 11   | F   | 6       | L    | SNHL       | Idiopathic   | 70          |
| 12   | F   | 5       | R    | CHL        | S/p COM      | 40          |
| 13   | M   | 5       | L    | SNHL       | Idiopathic   | 55          |
| 14   | M   | 7       | R    | SNHL       | Idiopathic   | 65          |
| 15   | F   | 5       | L    | CHL        | Post-tympanoplasty | 45          |
| 16   | M   | 8       | R    | SNHL       | Idiopathic   | 70          |
| 17   | M   | 8       | R    | CHL        | S/p COM      | 40          |
| 18   | M   | 6       | R    | SNHL       | Idiopathic   | 70          |
| 19   | M   | 5       | L    | SNHL       | Idiopathic   | 65          |

M: male; F: female; R: right; L: left; HL: Hearing Loss; CHL: Conductive Hearing Loss; SNHL: Sensorineural Hearing Loss; S/p COM: Post-Suppurative Chronic Otitis Media; BC: bone conduction.

Medical Indication

**Congenital:** Atresia/ Microtia, Ossicular chain malformations, Syndromic Hearing Loss

**Acquired (temporary):** Suppurative Acute /Chronic Otitis Media, Tubaric Dysfunction

Figure 1. AAP and its components.
This test’s score was calculated by considering the number of correct answers on the total number of questions. Following Bisiacchi, the total number of questions was set to 9; also, the final score was a number between 1 and 6, where scores between 1 and 3 indicated a short-term memory deficit, and scores above 4 normal short-term memory function. Similarly to the WM function test, this test was chosen because it specifically measures short-term memory abilities in children of age between 5 and 8 years.

**Statistical Analysis**

For each test and each subject, the difference between scores recorded at T1 and T0 was calculated. The WM tests scores of the SSHL group across T0, T1, and those of the CG were compared using a one-way ANOVA test followed by a post-hoc analysis using the Holm-Bonferroni (HB) method. A one-way ANOVA test and post-hoc analyses were also used to assess if there was a statistically significant difference between the scores of subjects with different SSHL severities at T1. The short-term memory test scores were analyzed in a similar manner. For all the tests, the level of significance was set to 0.05. Statistical analysis was performed using Stata.

**RESULTS**

Among the subjects with SSHL, 5 presented with a mild form of SSHL, 6 with a moderate form of SSHL, and 8 with moderate-severe SSHL. Four children had a congenital form of SSHL, 2 suffered from post-tympanoplasty disease, 2 from post-suppurative otitis media, and 10 had an idiopathic SSHL (Table 1). All CG subjects displayed normal bilateral hearing function (Table 2).

**Working Memory**

In the subjects with SSHL, compared with the WM test scores at T0, the scores of the WM test at T1 displayed only minor changes in the quiet condition but greatly improved in the noise condition (Table 3). Statistical analysis found no statistically significant difference between the WM test scores collected in quiet at T0, T1, and those of the CG (ANOVA: p=0.1). Conversely, there was a statistically significant difference between the WM test scores collected in noise at T0, T1, and those of the CG (ANOVA: p<0.0001). Post-hoc analysis confirmed that there was a statistically significant difference between the scores collected at T0 and T1 (HB: p<0.01), the scores collected at T0 and those of the CG (HB: p<0.01), and the scores collected at T1 and those of the CG (HB: p<0.01). Additionally, there was no statistically significant difference between the scores of subjects with different SSHL severities at T1 (t-test: p=0.9).

**Short-Term Memory**

In the subjects with SSHL, the scores of the short-term memory test at T1 improved compared with the scores at T0 (see Table 4 for a summary of results). The average score was 2.8 (SD: 0.8; CI 95%: 2-4) at T0 and 3.7 (SD: 0.6; CI 95%: 3-5) at T1. There was a statistically significant difference between the scores collected at T0 and T1 and those of the CG (ANOVA: p<0.0001). Specifically, there was a difference between the scores at T0 and T1 (HB: p<0.01) and between the scores at T0 and those of the CG (HB: p<0.01). Furthermore, the scores at T1 were significantly different from those of the CG (HB: p<0.01) (Table 3). There was no statistically significant difference between the scores of the subjects with different SSHL severities at T1 (t-test: p=0.8).

### Table 2. Scores of WM and short-term memory test in the CG

| Patient | Sex | Age | Silence | Noise | Short-term Memory |
|---------|-----|-----|---------|-------|-------------------|
| 1       | F   | 6   | 100%    | 100%  | 5                 |
| 2       | M   | 5   | 100%    | 100%  | 4                 |
| 3       | F   | 6   | 100%    | 100%  | 5                 |
| 4       | F   | 5   | 100%    | 100%  | 5                 |
| 5       | M   | 7   | 100%    | 100%  | 6                 |
| 6       | F   | 8   | 100%    | 100%  | 6                 |
| 7       | F   | 8   | 100%    | 100%  | 6                 |
| 8       | M   | 7   | 100%    | 100%  | 6                 |
| 9       | M   | 8   | 100%    | 100%  | 6                 |
| 10      | M   | 7   | 100%    | 100%  | 5                 |
| 11      | M   | 6   | 100%    | 100%  | 4                 |
| 12      | F   | 5   | 100%    | 100%  | 5                 |
| 13      | F   | 6   | 100%    | 100%  | 5                 |
| 14      | M   | 5   | 100%    | 100%  | 6                 |
| 15      | M   | 8   | 100%    | 100%  | 6                 |

M: male; F: female
The main result of this study was that in pediatric subjects with SSHL, AAP significantly improved short-term memory function and WM function in the noise (but not in the quiet) condition. However, the performances of the SSHL subjects remained significantly lower than those of an age-matched group of healthy controls regardless of the severity of their hearing impairment.

In the noise condition, WM test performances of SSHL subjects at T1 were overall and significantly improved compared with performances at T0. Individual patient analysis showed that in the noise condition, WM test scores improved in 18 out of the 19 SSHL subjects (Table 3). This improvement might be due to different causes. It may be the consequence of an improvement in speech discrimination (i.e., the ability to correctly identify vowels and consonants) possibly elicited by the AAP. Although our study did not specifically test subjects’ speech discrimination abilities, this hypothesis is consistent with the results of Di Stadio et al.[10] study that showed that a bone anchored hearing aid (BAHI) improved SSHL subjects’ ability to discriminate words both in silence and noise as well as memory function.[10] This hypothesis is also consistent with the results of Pisoni [14], who showed that the ability to correctly identify a word’s formants correlates with good memory function in subjects with normal hearing [14]. The WM improvement we observed in the noise condition might also be explained as a by-product of neural changes underlying the restoration of bilateral hearing function via AAP. Cortical neuroplastic changes/development [14, 15] might have been elicited through a mechanism similar to the one described by Sharma et al. in SSHL children with cochlear implant (CI) [23,24,25]. Another facilitation to memory function might have stemmed from a strengthening (possibly duplication) of the auditory signal arriving into the hearing cortex through squelch effect [26].

As shown in Table 3, one SSHL subject displayed a worsening in the WM test scores in the noise condition. This could be due to impairment of the subject’s ability to focus, which could be due to an increase in external hearing stimuli experienced after bilateral hearing recovery or [27] stress [27].

Statistical comparison showed that at T0 and in the noise condition, the WM test scores of the SSHL subjects at T1 were overall and significantly improved compared with performances at T0. Individual patient analysis showed that in the noise condition, WM test scores improved in 18 out of the 19 SSHL subjects (Table 3). This improvement might be due to different causes. It may be the consequence of an improvement in speech discrimination (i.e., the ability to correctly identify vowels and consonants) possibly elicited by the AAP. Although our study did not specifically test subjects’ speech discrimination abilities, this hypothesis is consistent with the results of Di Stadio et al.[10] study that showed that a bone anchored hearing aid (BAHI) improved SSHL subjects’ ability to discriminate words both in silence and noise as well as memory function.[10] This hypothesis is also consistent with the results of Pisoni [14], who showed that the ability to correctly identify a word’s formants correlates with good memory function in subjects with normal hearing [14]. The WM improvement we observed in the noise condition might also be explained as a by-product of neural changes underlying the restoration of bilateral hearing function via AAP. Cortical neuroplastic changes/development [14, 15] might have been elicited through a mechanism similar to the one described by Sharma et al. in SSHL children with cochlear implant (CI) [23,24,25]. Another facilitation to memory function might have stemmed from a strengthening (possibly duplication) of the auditory signal arriving into the hearing cortex through squelch effect [26].

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Statistical comparison showed that at T0 and in the noise condition, the WM test scores of the SSHL subjects were significantly different from the WM test scores of the CG; at T1 there was still a significant difference between the WM test scores of the SSHL subjects and those of the CG. Thus, although AAP improved their performances, SSHL subjects did not fully regain normal WM function in the noise condition as measured by our WM test. We speculate that this result reflects a delay in the recovery of WM function, possibly related to a difficulty of SSHL subjects in quickly adapting/re-adapting to bilateral hearing. Further studies with longer follow-ups (for example, 6 months or more) might help test this hypothesis.[16].

In the quiet condition, the performances of the SSHL subjects in the WM test did not overall improve at T1 compared with T0. This result

| Table 3. Results of WM tests at T0 and T1 in quiet and noise |
|-----------------|----------------|----------------|----------------|
| Working Memory  | Quiet          | Noise          | Working Memory  |
|                 | T0             | T1             | T0             | T1             |
| Patient         | 1              | 2              | 3              | 4              | 5              | 6              | 7              | 8              | 9              | 10             | 11             | 12             | 13             | 14             | 15             | 16             | 17             | 18             | 19             |
| 1               | 95%            | 100%           | 5%             | 50%            |
| 2               | 79%            | 100%           | 43%            | 95%            |
| 3               | 98%            | 100%           | 25%            | 75%            |
| 4               | 92%            | 15%            | 100%           | 15%            |
| 5               | 94%            | 100%           | 5%             | 50%            |
| 6               | 60%            | 50%            | 0%             | 30%            |
| 7               | 100%           | 93%            | 77%            | 72%            |
| 8               | 58%            | 100%           | 28%            | 90%            |
| 9               | 0%             | 0%             | 100%           | 70%            |
| 10              | 97%            | 100%           | 35%            | 90%            |
| 11              | 98%            | 100%           | 25%            | 90%            |
| 12              | 98%            | 100%           | 25%            | 75%            |
| 13              | 98%            | 100%           | 25%            | 90%            |
| 14              | 92%            | 100%           | 15%            | 80%            |
| 15              | 100%           | 100%           | 35%            | 90%            |
| 16              | 100%           | 100%           | 40%            | 95%            |
| 17              | 98%            | 100%           | 30%            | 95%            |
| 18              | 93%            | 100%           | 20%            | 80%            |
| 19              |                |                |                |                |

| Table 4. Results of short-term memory tests at T0 and T1 in quiet |
|-----------------|----------------|----------------|
| Short-term Memory (Quiet) | Patient | T0 | T1 |
|                 | 1         | 4  | 5  |
|                 | 2         | 3  | 4  |
|                 | 3         | 3  | 4  |
|                 | 4         | 4  | 4  |
|                 | 5         | 2  | 3  |
|                 | 6         | 4  | 4  |
|                 | 7         | 4  | 4  |
|                 | 8         | 2  | 4  |
|                 | 9         | 3  | 3  |
|                 | 10        | 3  | 4  |
|                 | 11        | 3  | 4  |
|                 | 12        | 2  | 3  |
|                 | 13        | 2  | 3  |
|                 | 14        | 3  | 4  |
|                 | 15        | 2  | 3  |
|                 | 16        | 3  | 4  |
|                 | 17        | 3  | 4  |
|                 | 18        | 2  | 3  |
|                 | 19        | 2  | 3  |

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could be due the fact that the SSHL subjects’ WM test scores at T0 were almost normal (Table 3). Additionally, 2 SSHL subjects displayed worsened performance (patient 6 and 7). Normal variability in performance on the day of testing might account for this worsening. In SSHL subjects, short-term memory function significantly improved after 1 month of use of AAP. However, none of the SSHL subjects achieved a score of 6 (i.e., a normal score) at T1. As discussed above, future studies with longer follow-ups might allow us to test whether AAP can help children with SSHL achieve performances comparable to those of healthy controls. In this patient population, times longer than 1 month (e.g., 6–12 months) might be necessary for the AAP to induce significant neuroplastic changes. Longer follow-ups might also allow us to investigate inter-subject variability in recovery times. Additionally, larger sample sizes and functional MRI data might help elucidate the effects of bilateral hearing function restoration on brain plasticity.

Although previous studies have shown that the use of AAP is only indicated in subjects with a hearing threshold equal to or lower than 25 dB (only in these cases a normal hearing threshold can be restored), we observed that AAP improved WM and short-term memory functions in subjects with conductive single side hearing loss (CSSHL) with hearing thresholds greater than 25 dB. This result suggests that even a partial recovery of hearing function in the ear suffering from hearing loss can lead to memory function improvement, which is not surprising, because bilateral hearing function is key for preserving memory function and verbal and word recollection abilities. A possible underlying mechanism for such improvements after binaural hearing function restoration is the squelch effect, which, through a more efficient signal integration into the brain, enhances the subjects’ ability to identify and recall sounds in the voice frequency band and correctly discriminate words, especially in noisy settings. On the basis of the results of these previous studies, which are consistent with those presented herein, our group has long been advocating bilateral hearing function restoration in (both pediatric and adult) patients with SSHL, regardless of disease severity and cause.

In a previous study, Di Stadio et al. showed that in children with SSD treated with a BAHI, memory performance was similar to that of healthy, age-matched controls. The results presented herein are consistent with the results by Di Stadio et al. and show that restoring bilateral function in children with SSHL has a positive impact on memory function. Taken together with the results of the studies that have shown that even a mild hearing function loss might cause cognitive fatigue and negatively impact subjects’ intellectual abilities and academic performance, the results of these studies suggest that in children with unilateral hearing impairment, bilateral hearing should be restored regardless of the impairment severity. This idea is supported by the study of Stiles et al. on children with bilateral hearing loss treated with a bilateral hearing aid. Conversely, other studies on deaf children suggest that bilateral hearing restoration through CI may increase the risk of neurocognitive decline or delay development of WM capacities. Although the findings of these studies should be interpreted with caution (a comparison between results of studies where hearing aids are used to treat patients with unilateral and bilateral hearing impairments is difficult, and studies by Nittroeur et al., lack a control group) or are at odds with those of the studies in older adults that have shown instead that bilateral hearing restoration decreases the risk of cognitive decline, they highlight limitations of current knowledge on the effect of different types of hearing aids on memory function in children and the need for further, systematic studies. Given the potential impact of memory function on academic performance, gaining such knowledge might not only be helpful to clinicians for the planning of treatments but also to society.

Because it does not require surgical implantation, AAP is an ideal tool for investigating whether and to what extent restoration of bilateral hearing function impacts cognitive development in children with unilateral hearing impairment. Children could undergo a “trial” period with the system (e.g., 6 months), during which cognitive testing could be performed in order to assess potential cognitive decline; at the end of such trial period, the collected data could be used to guide the most appropriate course of treatment (e.g., proceeding with CI surgery or keeping the AAP). In 2015 Arndt et al. showed that 50% of children with SSD displayed a hypoplasia/aplasia of the cochlear nerve as shown by MRI, suggesting that in these patients, an AAP might be a better option than a CI.

Additional studies evaluating the effect of traditional hearing aids (HA) on memory function should be performed. Furthermore, whether different hearing restoration systems such as HA, BAHI, AAP, or CI differentially impact memory abilities in children with different WM or short-term memory test scores should be investigated.

Study Limitations
The main limitation of this study is the relatively short follow-up (1 month). Longer follow-ups may show improvement in memory functions greater than those we observed. Studies on a wider sample, where subjects are stratified by age, should also be performed.

CONCLUSION
To the best of our knowledge, this is the first study that investigated the effects of AAP on WM and short-term memory functions in children with SSHL. We found that in this patient population, restoration of bilateral hearing function with AAP improved patient performance in WM function tests in noise and in short-term memory function tests. However, patients’ scores remained lower than those of an age-matched CG, possibly because the relatively short follow-up implemented in this study. We recommend that AAP be used for non-invasive treatment of SSHL and that AAP be worn as long as possible in order to improve all memory functions.

Ethics Committee Approval: Ethics committee approval was received for this study from the Internal Review Board of Santobono-Possilippo Hospital in Naples.

Informed Consent: Written informed consent was obtained from the patients who participated in this study.

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