Preservation of Singing Functioning in a 5 Year-Old Following Severe Right-Sided Traumatic Brain Injury: Insights into the Neurological Resilience of Song from Pediatric Music Therapy

Ellen C. Gentle¹, Melinda Barker¹, Janeen Bower¹
¹The Royal Children’s Hospital, Melbourne, Australia

Abstract
Studies examining song functioning in childhood are of particular importance when devising developmentally appropriate evidence-based Music Therapy (MT) interventions during recovery from brain injury. In comparison to adult studies where neural organization may be well defined, the neural organization of song in the developing brain has been under-researched. This includes functional consequences following neurological insult. This case study documents a 5 year-old female with typically developing language and verbal memory that suffered a severe traumatic brain injury. Despite extensive right hemisphere damage, her recognition and memory of previously well-learned (familiar) songs was preserved. New learning and retention of unfamiliar songs with lyrics was also observed and was not predicted based on adult models of melodic learning. Findings suggest that the song system in childhood is a neurologically significant, robust system not easily disrupted following extensive brain injury, and caution against assuming adult models of music organisation in the developing brain.

Keywords: song development, child development, music therapy, childhood brain injury

Introduction
Song is a vital part of early childhood development. In infancy the human brain demonstrates an innate ability to attend to and process both melodic and linguistic aspects of the auditory environment [1]. Infants are able to respond to, and produce song patterns even before other higher order cognitive skills, like language become discernable [2,3]. The acquisition of the song lexicon occurs similarly to the language lexicon through enculturation and exposure to melody and lyrics combined in song [4]. Early Western musical experiences are typically culturally specific songs with words such as lullabies and nursery rhymes. Although the ability to recognize, understand and produce familiar songs are an integral part of most western childhood cultures, there is a paucity of research focusing on the neural development and functional organization of this skill.

Research in non-musically trained adult populations tells us that song has a close relationship with language in the adult brain, with recent imaging studies demonstrating that the processing and production of singing and language uses overlapping networks of bilateral, predominantly front-temporal neural activity with singing more strongly lateralized to the non-dominant hemisphere (the right hemisphere in typically developed adults [5]), and language to the dominant left hemisphere [5-9]. When initially learning a song with words, adult models suggest various levels of integration of music and language in the brain [10], with both shared and separate neural substrates for the song learning process [11]. For the melodic aspects of song, typically developed adults show a strong right hemispheric bias for both semantic and episodic memory [12]. Memory structures such as the right hippocampus and surrounding mesial temporal lobe are strongly involved in musical memory tasks [13] and are vital to the acquisition of the tune lexicon and the formation of song memory [14]. When a song has words, a significant contribution to learning has also been evidenced from the left hemisphere, with a posterior-anterior gradient of integration along the left superior temporal gyrus/sulcus for lyrics and tunes observed [10]. Recent functional Magnetic Resonance-adaptation findings also indicate that when healthy adults learn a song with words, learning occurs most efficiently and more robustly where the language-dominant hemisphere is involved [15]. Thus the ability to listen to a song, recognize it,
learn it, become familiar with it, commit it to memory and be able to sing it with words requires a complex network of bi-hemispheric brain regions.

Understanding the neural basis of singing is particularly important for individuals using music-based therapies for rehabilitation, where estimating capacity of musical functioning following brain damage is vital to the efficacy of the interaction. In current Music Therapy (MT) practice, Neurologic Music Therapy (NMT) is often used when working with individuals with an acquired brain injury. It is a systematic, evidence-based model seated on principles of adult neural organization which utilizes music interventions to remediate non-musical disorders or deficits that result from brain injury or disease [16]. When applying adult models to children in order to understand song functioning however, a number of difficulties arise. In the developing brain lateralisaton of hemispheric functions are not as well established as they are in adults. Caution is often advised when using adult models of hemispheric organisation and lateralisaton to estimate brain correlates of functioning in children [17]. Functional organisation of the developing brain is even less predictable in the context of early brain damage [17]. There is a growing incentive for those who work musically with children recovering from a neurological insult to have access to developmentally relevant models of music organisation.

Music Therapists in pediatric acquired brain injury populations, typically use songs as the main medium to stimulate physiological and behavioral responses [18,19] and to reduce agitation [20] in children emerging from coma. The temporal predictability of a highly familiar song is also thought to provide an external focus of stability and emotional comfort to lessen the impact of neurological disturbance for children in a coma [21]. The precise neural mechanisms behind the efficacy of these types of musical intervention remain largely unexplored however. In theory, the extensive bilateral nature of the song network may increases the potential for retaining functionality in the presence of neural damage compared to other, more circumscribed networks of function [21]. Thus, in cases of severe brain injury, very familiar songs may be able to stimulate meaningful responses that support early recovery in a way that verbal stimuli (language) and other sensory stimuli may not [21,22]. While this premise has not been tested in children, case studies of adults with brain damage would suggest that familiar songs with words in particular have the potential to be neurologically accessible following brain injury. Adults with extensive right-hemisphere damage resulting in amusia (an acquired deficit in musical knowledge including recognition), with preserved verbal abilities, have demonstrated that familiar song-based musical knowledge can still be accessed in some way, presumably because of an over-learned association with lyrics (e.g. ‘Happy Birthday’). Over-learning of songs with words may therefore represent a special form of neural representation which is more resistant to interference post brain injury [23-25].

Currently, there are no reported case studies that investigate the significance or organisation of song functioning in children with an acquired brain injury. Examining a developmentally significant musical function such as singing after injury in childhood is needed. Not only to inform our understanding of musical organisation in the developing brain, but also to inform clinical practices during MT interventions to maximise outcomes. Studying such phenomena in vivo with such a vulnerable patient population is difficult due a variety of heterogeneous factors including nature of injury, age at insult and levels of parental distress. Behavioral observations during standard clinical practice MT sessions provide a novel and non-invasive approach to study the nature and trajectory of recovery of musical functions after childhood brain injury. The following case represents a pivotal step toward understanding singing functioning in the developing brain and highlights the role that suitably trained music therapists might use in furthering our understanding of musical functioning in the presence of neurological damage.

Methods

Case study: Patient Helen

‘Helen’ was a 5-year and 7 month-old girl who sustained a severe right-hemisphere brain injury after being struck by a motor vehicle travelling at approximately 50km/h. At the time of injury Helen was unconscious with a fluctuating Glasgow Coma Scale (GCS) of 3-5. On admittance to hospital Computerised Topography (CT) and Magnetic Resonance Image (MRI) scans revealed extensive right-sided damage which included a large temporoparietal depressed skull fracture involving the skull base, sub and extra dural haemorrhage and associated midline shift, multiple frontal haemorrhagic foci and significant oedema plus fractures of the medial maxillary sinus and orbital haematoma. Surgical intervention 24 hours post-injury included a right craniecctomy, a right subtemporal decompression and insertion of an intracranial pressure (ICP) monitor. Following surgery, herniation of the brain through the cranieotomy site necessitated an extension of the cranieotomy and further right frontal-parietal partial lobectomy. At 72 hours post-injury Helen underwent further surgery to widen the bone flap and further dissect the herniating brain in the parietal and frontal-temporal regions. The extent of Helen’s brain damage, particularly within the right temporal lobe at 7 days post-injury can be seen in Figure 1. While her left hemisphere was relatively preserved, there was evidence of slight ischemic change on MRI within the left frontal lobe, posterior cingulate gyrus and subcortical white matter.
MT with Helen commenced 26 days post-injury when she was medically stable and self-ventilating. At the outset of the MT service, Helen’s mother provided a list of H’s pre-morbid musical preferences, which largely included culturally specific nursery rhymes with simple intervals such as 3rd’s and 5th’s, such as ‘Twinkle, Twinkle Little Star’ and ‘Baa-Baa Black Sheep.’ Helen’s mother reported that Helen had been familiar with these songs since infancy and had regular exposure to both listening and singing them prior to her accident. The MT program was conducted by an experienced registered MT with NMT training. The primary approaches to the therapy are detailed below and were NMT-based, selected to maximise recovery systematically while supporting Helen’s emerging cognitive, physical, language and emotional rehabilitative needs. Song was the primary medium of musical engagement throughout MT sessions.

**Musical Neglect Training (MNT) & Music Attention and Control Training (MACT)** [16]

Helen presented with left-sided neglect, a dense left hemiplegia, and difficulties with sustained and divided attention. MNT was used to support Helen’s attendance to her neglected side by encouraging active performance of musical instruments that were spatially configured in Helen’s left (neglected) visual field. Musical cues, including rhythm and melody were structured to provide auditory cues to support Helen’s playing of instruments. For example, a descending melody line given by the music therapist provided an auditory cue for Helen to follow the melody on her xylophone with the lower register placed in her neglected visual field. MACT was used to promote sustained and divided attention during recovery. This comprised musical activities where the music therapist manipulated different musical sounds/instruments to engage Helen’s attention, and help her to focus, sustain and divide her attention for increasing periods.

**Musical Sensory Orientation Training (MSOT)** [16]

During MSOT music was presented live, with pre-morbidly identified familiar songs used extensively to stimulate arousal, awareness and orientation, and facilitate meaningful responses and engagement. Sessions typically began with a greeting- ‘Hello Song’, which repetitively used Helen’s name to promote a sense of routine and increase orientation. Pre-morbidly familiar songs were then sung with a simple guitar accompaniment to facilitate intentional responses. As Helen’s recovery progressed, the length of the MSOT was extended and she was encouraged to engage more actively in simple musical activities to promote responsiveness, basic attention maintenance and engagement. This included encouraging Helen to join in singing familiar songs.

*Table 1* details the key components of Helen’s recovery throughout her MT sessions, including the techniques used, content of the sessions and the responses/behaviors observed during sessions with Helen.

**Results**

*Table 1* details the remarkable progression of functional singing recovery for Helen, which can be seen to largely parallel her language recovery, amnestic phase and ability to attend and engage for longer periods of time in sessions. At just over 6 weeks post-injury, she demonstrated obvious awareness of familiar songs given by her positive behavioural responses during expectancy pauses. At 8 weeks post-injury, not only was Helen able to speak the words of pre-morbidly familiar songs, she did so with little repetition and showed increasing melodic contour in her singing. By the time Helen emerged from the PTA phase at just 12 weeks post injury she was able to tunefully sing along with pre-morbidly familiar songs. During the post-PTA phase she demonstrated recognition of pre-morbidly familiar songs (nursery rhymes) based solely on their melody. Helen was also then able to sing these songs without musical accompaniment or prompting from the music therapist tunefully. Preliminary investigation
suggested her musical working memory was also intact as indicated by her ability to retain and repeat short unfamiliar melodic phrases. The most unexpected finding during recovery however, was Helen’s demonstrated new learning and recall of previously unfamiliar songs with lyrics. This finding became apparent when Helen’s mother began to sing a popular rock song to her during their leisure time in the hospital. Helen was then able to spontaneously recall this song.

Table 1. Timeline of H’s inpatient recovery, Music Therapy (MT) intervention sessions and behavioral responses

<table>
<thead>
<tr>
<th>MT Session</th>
<th>Time since injury</th>
<th>Medical status</th>
<th>Functional recovery stage</th>
<th>Session content</th>
<th>Observed behaviours and responses</th>
</tr>
</thead>
<tbody>
<tr>
<td>1-3</td>
<td>26-33 days</td>
<td>Post Traumatic Amnesia (PTA)</td>
<td>Periods of wakefulness with impaired awareness, some confusion No verbalisations due to tracheostomy</td>
<td>MSOT Favourite nursery rhymes sung live with simple guitar accompaniment songs included ‘Twinkle twinkle little star’, ‘Five little ducks’, ‘Open shut them’</td>
<td>No observed behavioural responses to music. In later sessions began visually orienting to right side. Completed hand actions to ‘Twinkle twinkle little star’ with right hand</td>
</tr>
<tr>
<td>3-5</td>
<td>36 – 43 days</td>
<td>PTA</td>
<td>Impaired attention span and poor concentration Purposeful movement on right side Able to obey basic commands</td>
<td>MSOT extended for increasingly longer periods of time</td>
<td>Good eye contact. Smiled in response to singing her name. Completed hand actions to a variety of pre-morbidly learned nursery rhymes with right hand</td>
</tr>
<tr>
<td>6</td>
<td>47 days</td>
<td>PTA</td>
<td>Impaired attention span and poor concentration Increased movements on right side Spontaneous noises verbalised</td>
<td>As above plus stimulation of automatic verbalisations: therapist left space in song lyrics to ‘Twinkle twinkle little star’</td>
<td>No verbalisations during song but consistently smiled during spaces indicating awareness of an expectation for her to respond</td>
</tr>
<tr>
<td>7-9</td>
<td>48-57 days</td>
<td>PTA</td>
<td>More alert and oriented Improving attention span and variable concentration Limited vocabulary uttered, echolalia present.</td>
<td>As above plus H was verbally encouraged by music therapist to sing along.</td>
<td>Able to participate in song: Initially talked song lyrics mono-tonally but after several repetitions began singing along with music therapist with correct lyrics and relative melodic contours.</td>
</tr>
<tr>
<td>10+</td>
<td>8-12 weeks</td>
<td>PTA</td>
<td>Alert and oriented Improving attention span and variable concentration Talking in very short sentences, some echolalia present.</td>
<td>As above plus playing of small hand held percussion instruments introduced</td>
<td>Continued to sing along with a greater number of nursery rhymes that were pre-morbidly familiar. Concentration maintained for song duration (up to 90 seconds)</td>
</tr>
<tr>
<td>20+</td>
<td>12+ weeks</td>
<td>No PTA</td>
<td>Improving attention span and variable concentration Increased use of language (sentences)</td>
<td>MNT &amp; MACT introduced (30-60 minutes) plus introduction of new song material (unfamiliar songs) including: songs with lyrics and melody only phrases.</td>
<td>Able to: i) name familiar (previously learned) nursery rhymes when melody only presented ii) repeat short melodic phrase (4-6 notes) back to music therapist. iii) learn a new song with lyrics outside of MT sessions with parent and then sing along with music therapist iv) sing a new song with lyrics with tuneful melodic contour, with only guitar accompaniment, some prompting for lyrics required</td>
</tr>
</tbody>
</table>

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to the music therapist after some time and was able to correctly produce the chorus, with a relative melodic contour and only guitar accompaniment. It was also observed that singing the melody of the unfamiliar new rock song was not difficult for Helen, but remembering the lyrics paired with it was, and she required prompting for this at times.

**Additional neuropsychological results**

A neuropsychological assessment of Helen at 6 years and 6 months of age, 11 months post-injury, estimated her pre-morbid IQ to be within *Average* ranges. Some residual left hemiplegia was noted. Tests revealed broadly intact verbal skills, which suggested language development was typically left-hemispherically located. Concordant with the site of her injury, Helen’s visually based skills were significantly impaired; falling well below age expected levels. Her new learning and memory for both visual and verbal information was not significantly impaired, but impacted upon by impaired information processing, poor attention control and limited working memory skills.

**Discussion**

The case of Helen illustrates the potential for neurological resilience within the song network in children as young as 5 years of age. Helen demonstrated a remarkable retention of song-based functionality post-injury that was unexpected based on current adult models of musical functioning. Helen’s retention of pre-morbidly over-learned songs with words (nursery rhymes) did support adult findings suggesting that for overly familiar songs with words, the pairing of melody and lyrics may represent a special form of learning more resistant to right hemisphere damage [15]. Not predicted by adult models of neural organisation for music [23,25,26] was the finding that post-PTA, Helen showed no obvious impairment in her recognition of previously learned songs, musical working memory nor her ability to acquire, retain and sing with relative melodic contour new songs with words. Given the extensive damage throughout her right hemisphere, including the mesial temporal lobe, this finding pointed strongly to left-hemisphere involvement in new melodic learning for songs with words.

It was also observed that recalling the melody of the unfamiliar new rock song was not difficult for Helen, but remembering the lyrics paired with it was and she required prompting for this. Given that Helen was still in neural recovery it is unsurprising that integration of melodic and verbal information into memory was still challenging for her. This finding may also point to the decreased efficacy of unilateral encoding for songs where previously bilateral encoding had occurred [10,15]. Unfortunately due to her age and ongoing attention and behavior impairments Helen could not be scanned using functional imaging techniques to examine the extent or overlap of left hemisphere brain regions involved in her singing and language functioning post-injury. Helen was just over 5 years of age at the time of injury. Neuroimaging studies suggest that critical periods of lateralisation for language occur around this age (4-5 years) [27,28] and this process continues in coordination with the consolidation of language development [28]. Intact speech and language functioning post-injury and neuropsychological testing confirmed that Helen’s verbal skills and language development were most likely lateralised to the left hemisphere pre-morbidly and were typically developing. This suggests that Helen was using her linguistic hemisphere to sing melodically and to learn new songs, and raises questions such as whether i) the neural substrates of music, like language, follow similar neuro-developmental trajectories of lateralisation and ii) if Helen’s retained musical abilities were contributed to by greater availability of shared neural resources post-injury. At this time far more research is required to explore the early neuro-developmental organization of music, singing and language to support these theories. Regardless of the mechanism of recovery however, her case demonstrates the scope for recovery of musical functions, and in particular singing in early childhood brain injury.

With regards to paediatric clinical MT practice, the findings from this case study have significant implications, particularly for therapists using song during neural recovery. Helen demonstrated that despite severe and extensive brain injury, young children can have the capacity to engage meaningfully, show new learning and thrive in song-based therapy. For music therapists using song in pediatric acquired brain injury populations, level of song familiarity, age at injury and language markers may become increasingly important considerations in therapy planning and predicting recovery outcomes. Lastly, the case of Helen once again provides a cautionary tale for those assuming adult models of brain organisation to inform pediatric practices.

**Conclusions**

This case represents a significant step forward in increasing the evidence base within developmental music neuroscience, and highlights the capacity for dynamic musical organisation in the developing brain. It also demonstrates that pairing MT with neuroscience can greatly facilitate understanding of the neurological basis of music, and in particular the neurological significance and rehabilitative potential of song within the developing brain. Finally, this article calls for more case studies of this type that aim to characterize recovery of musical functioning in the context of developmental brain injury.
References


Biographical Statements

Dr Ellen Gentle, DPsych (Clin Neuro), is a Clinical Neuropsychologist who provides neuropsychological assessment, education and intervention for children aged 4-18 diagnosed with a range of neurodevelopmental, learning and acquired disorders of childhood and adolescence within the psychology department of The Royal Children’s Hospital, Melbourne.

Dr Melinda Barker, DPsych (Neuro), is a Senior Clinical Neuropsychologist who provides inpatient and outpatient neuropsychological services including assessment, education and behaviour management support to families of children who have acquired brain injuries.

Janeen Bower, MMus(MusThryp), NMT, RMT, is a Clinical Music Therapist in the Neuroscience and Rehabilitation Departments at The Royal Children’s Hospital Melbourne, Australia.