

Possible Implications of a Shunt Surgery on a Blind Child's Academic and Spatial Orientations Skills - A Case Report

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Abstract

In various cases of visual impairments, the individuals are referred to expert Ophthalmologists in order to establish a correct diagnosis. Children with visual-impairments confront various challenging experiences in life since early childhood throughout lifespan. In some cases, blind infants, especially due to congenital hydrocephalus, suffer from high intra-cranial pressure and, consequently, go through a ventriculo-peritoneal shunt surgery in order to limit the neurological symptoms or decrease the cognitive impairments. In this article, a detailed description of numerous crucial implications of the V/P shunt surgery, through the right posterior-inferior parieto-temporal cortex, on the observed preliminary capabilities that are pre-requisites for the acquisition of literacy skills in braille, basic Math competencies, braille printing which suggest Gerstmann syndrome in the blind. In addition, significant difficulties orientation and mobility skills using the Cane, in general organizational skills and social interactions were observed. The primary conclusion of this report focuses on raising awareness among neuro-surgeons towards the need for alternative intracranial routes for V/P shunt implantation in blind infants that preserve the right posterior-inferior parieto-temporal cortex that is hypothesized to modulate the tactual-spatial cues in braille discrimination. A second conclusion targets educators and therapists that address the acquired dysfunctions in blind individuals due to V/P shunt surgeries.

Keywords: *Blindness; Shunt implantation; Spatial orientation; Mobility skills; Literacy skills; Right Hemisphere*

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Introduction

Visual impairments or total blindness can be caused by various factors in infants, children and adults. In such conditions, the individual is referred to an Ophthalmologist in order to determine the correct diagnosis of each case.

The basic ophthalmological assessment, which can be done in the private practice, comprises of visual acuity measurements with correction, the Amsler Grid test (test with squares) for determining metamorphopsia, an examination of the anterior segment with a slit lamp, a gonioscopy for early diagnosis of new vessels in the angle between the iris and the cornea, a tonometry (measurement of intraocular pressure), a fundoscopy by dilated pupils of eyes the doctor is able to examine the optic disc, the macula, vitreous and retinal periphery and a digital photograph, if feasible. The photograph of the fundus records the patient's clinical presentation and in doing so we can monitor any progression of the disease (American Academy of Ophthalmology, 2016).

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School-aged children with severe visual impairment or congenital-blindness, without additional disabilities invest intensive efforts in refining their tactual skills and enhancing their perceptual abilities in order to gain proficient braille reading and Literacy. Braille code learning requires spatial-perceptual skills and cognitive strategies related to the various components of the braille stimuli. As academic tasks become more complex and abstract, these children cope with novel skills that require motivation, attention and concentration, visual imagery, proficient consolidation of auditory memory and rich lexicon (Jarjoura, 2004). These capabilities can be achieved in case that the children have intact neuroanatomical and neurophysiological functions. In some cases, children with visual impairments are born with life-threatening conditions that require surgical interventions in order to prevent additional dysfunctions and save their lives, such as Ventriculo-Peritoneal (V/P) shunt surgeries that aim to prevent complications of Hydrocephalus due to an increase in the intracranial pressure (ICP) caused by an uncontrolled increase in the creation of the Cerebro-Spinal Fluid (Sherman & Wensheng, 2008).

Although V/P shunt surgery is considered as a vital, life-saving surgical procedure (Lutz, Venkataraman & Browd, 2013), in visually-impaired children it may pose specific challenges in the shape a profound dysfunctions in spatial orientation, spatial perception and tactile word reading in braille code if the shunt surgery is repeated again due to a failure in the first shunt functions and, specifically, if the second shunt is penetrating through the right posterior-inferior parieto-temporo-occipital cortex and the intra-parietal sulcus in the right hemisphere (Siuda-Krzywicka, Jednorog, Marchewka & Amedi, 2016). Previous studies early up to the 80's of the 20th century showed even that V/P shunt surgeries do not achieve highly-efficient outcomes in preventing complications of Hydrocephalus in preterm infants (Boynton, Boynton, Merritt, Yvonne, James & Bejar, 1986). The same authors stated that "...a total of 34 infants (68%) required one shunt revision or more, and the overall infection rate per patient was 50%. Seven infants died, 2 from shunt infections". Regarding visual, audiological, ophthalmological and neurodevelopmental assessments' outcomes, the authors stated: "...of the survivors, 11 (28%) have severe visual loss and 10 (24%) have hearing impairment. Of the infants, 21 (49%) have severe motor handicaps and 19 (38%) have seizure disorders".

Other complications of the V/P shunt malfunctions can be found in Arroyo, Jan, McCormick & Farrell (1985). The authors stated that: "...Fourteen (1.8%) of more than 800 children evaluated in a Visually Impaired Program over 10 years became permanently blind during an episode of raised intracranial pressure secondary to shunt malfunction. Clinical and radiologic findings suggested that the loss of sight was caused by a lesion in the pregeniculate pathway in nine patients, probably due to ischemia; five children that had surgeries in the postgeniculate cortex sustained infarcts of the occipital lobes".

Failures of V/P shunt surgeries were documented also in other cases of health conditions such as in Neurocysticercosis that leads to seizures. Singhi & Singhi (2009) reported that twenty-one patients required 49 operations for relief of Hydrocephalus, including 22 new shunts placements, 23 revisions and 4 cyst extirpations. Of these shunt failures, 78% occurred within that first 12 months and 96% within 3 years. Of the failures occurred in the distal segment, 75% occurred within 6 months. By comparison, 33% of proximal segment obstruction and 50% of the total number of valve obstructions occurred within the first 6 months. Also Nfonsam, Chand, Roseblatt, Turner & Luciano (2008) reported that all 13 patients in their sample needed Laparoscopic management in order to relieve the related clinical symptoms of distal shunt complications.

Even sterile shunt with a silicone implant and proteins and polymeric substances were found to evoke immune-responses that may play a significant role behind sterile shunts' malfunctions (VandeVord, Gupta, Wilson, Vinuya, Schaefer, Canady & Wooley, 2004).

Few studies reported a recovery of sight after a shunt revision due to malfunctions that caused an increase in the intracranial pressure (Constantini, Umansky, Nasser & Shalit, 1987; Connolly, Jan & Cochrane, 1991).

The following single case-study described in this article aims to discuss the impacts of a V/P shunt surgery on AR's academic achievement, Braille reading, Arithmetic and numerical calculations and spatial orientation. Ophthalmological considerations will be discussed in the relevant parts.

A Case Report

AR was born on 2-1-2004 and as a premature infant (27 weeks, 940 grams), APGAR score 1/4. Placental Hemorrhage and respiratory distress syndrome of the newborn affected AR's fragile health condition. He was diagnosed with Intra-cranial pressure due to: [1] Intra-Ventricular Hemorrhage of the newborn, Grade III, Right Hemisphere, [2] Intra-Ventricular Hemorrhage of the newborn, Grade IV, Left Hemisphere, which required a surgical intervention for V/P shunt implantation through the right posterior Parieto-Temporal cortices and associative areas connecting the cortex with sub-cortical structures and the right lateral ventricle (Medical report, 20-6-2010).

AR was acknowledged by the committee of the National Insurance Institution as a legally-blind child and eligible for all services for the visually-impaired individuals in Israel. Later, at age of three years-old, AR was referred on 1-9-2007 to a special education Kindergarten at the "Convent of Nazareth" Primary-school for the Blind and Deaf in Nazareth due to a delay in various developmental skills, primarily perceptual, cognitive and lingual, e.g., basic orientation and mobility skills inside the classroom and in the surrounding environment did not improve although an intensive and daily training program was applied. Moreover, according to the educational reports of the Kindergarten teacher dated on the 15th of November 2009, AR did not seem to improve his language pragmatic skills congruently with the relevant social contexts even-though he acquired a new lexicon and fair psycho-social interactions.

At age 6 years-old, on 20-6-2010, AR underwent another V/P shunt surgery for a revision of the previous shunt that has been infected due to Entero-Bacteria which caused obstructive Hydrocephalus. In this recurrent surgery, the first Catheter was not removed due to obstruction, thus leading to penetrating a second Catheter in an adjacent cortical area.

Literacy skills' acquisition

AR was enrolled in first grade on 1-9-2010 at the same special education school in order to acquire basic literacy skills in Arabic Braille, both in his mother-tongue (Arabic) and Math skills, in addition to self-care skills. In later years, AR was engaged in other disciplines such as Arts, Islamic Religion, Basic Science, second language (Hebrew). Although standardized, valid and reliable methods of Braille teaching were applied for, at least, three years, AR did not succeed to tactually discriminate nor understand the various embossed Braille templates as compared to the basic Braille cell that constitutes of an embossed six-dot matrix. Thus, AR could not establish the initial association between each tactual Braille template and its congruent phonological representation in Arabic Alphabet in order to develop phonetic spelling and basic reading skills. Different alternative instructional approaches that were applied by the school's educators and multidisciplinary staff did not improve AR's insufficient alphabetical knowledge.

Here, it is assumed that the braille code discrimination and reading require intact posterior-inferior parieto-temporal cortices in order to discriminate and mentally-represent the tiny components of each single braille template according to its congruent sequential enumeration, to scan the sequential letters in each row, to differentiate between braille consonants, vowels and other tactual symbols such as punctuation marks, to detect errors and odds and to sustain orientation in the page being tactually-screened.

In parallel to the intensive training on foundational Braille spelling and reading, the basic Braille printing tasks were initiated in order to facilitate copying and printing skills but intensive repetitions and training of bilateral manual usage of the Perkins-Brailler did not result in proficient gains meaningful outcomes. The lack of a precise timing of performance and insufficient motor sequencing obviously contributed to a profound limitation in performing the bilateral printing tasks. Both skills are considered as preliminary pre-requisites for accurate and efficient Braille printing of the alphabet, digits, symbols and punctuation marks (Jarjoura, 2004, in Hebrew).

Activities of daily living

Activities of daily living (BADL's and IADL's) were also evaluated by structured and non-structured observations during daily routines at school. Currently, AR still does not succeed in manipulating his shirt in-space nor tying his shoelaces. In addition, He does not succeed to manipulate the belt into his trousers nor to button his shirt sleeves. These tasks, and other similar daily routines that are characterized by the prerequisites of spatial relations and orientation and motor planning seem to challenge AR's abilities and perceptual skills.

Orientation and mobility skills

Another functional domain that was significantly limited according to all assessments was the orientation and mobility skills and that are considered as vital and crucial for daily functions especially for individuals with visual impairments or total blindness (Bradley & Dunlop, 2005). AR was trained for three years by a qualified Orientation and Mobility (O&M) trainer both inside the school and in AR's residential environment. Although procedural learning (training and repetitions) and declarative knowledge (instructions, guidance and directions) were applied repeatedly and intensively, AR still need individual accompanying inside the school and at home. Actually, orientation at home was relatively easier than at school due to the various environmental cues and such as sounds and indoor activities of family members. In addition, the smaller and more restricted area at home actually supports AR's mobility and orientation skills during daily, more frequent functional activities.

Arithmetical milestones in Braille

A third functional domain that is considered as a crucial component of literacy skills was arithmetical milestones, in general, and in Braille, in particular. According to AR's 2013-2014 individual educational Program (IEP) he could not perceive nor internalize any basic number-related concepts. Even automatic serial count between 0-100 was limited and inaccurate. In addition, AR presented incapability to compare amounts, measures and numbers. Thus, addition, subtraction and multiplication were mathematical procedures that became beyond AR's academic, perceptual and cognitive learning-potential.

Finger count was intensively exercised and repeated through the first years at school by different educators, through various teaching strategies focusing on fingers' naming, sequence and counting in one hand and in both hand. Currently, at the age of 14, AR still does not succeed in counting his fingers in the accurate sequence nor showing the number of fingers that is equal to a number previously mentioned verbally or to the number of familiar items and objects. It seems that AR has the characteristics and signs of Acalculia of the spatial type which involves sequence and directions perception in addition to a distortion in orientation in the scheme of the palm and fingers.

The visual cortical areas and its intra-cortical regions contain associative areas that serve the spatial functions and the spatially-based procedural knowledge such as arithmetical procedures and numerical abilities (Nieder & Dehaene, 2009).

Basically, the association between each braille template's embossed dots and its spatial orientation and enumeration from 1-6 could not be established, regardless of the intensity of training and the number of repetitions (i.e., procedural learning). These outcome measures profoundly indicate a case of Acalculia (Denburg & Tranel, 2012), supposedly secondary to the neural lesion at the posterior-inferior parieto-temporal cortices due to the shunt's implantation. In other words, the consolidation of number representation in the cortex could be established.

Organizational Skills in Social and Physical Environments

AR's social competencies and interactions with peers and staff members were characterized by shallow understanding of social situations and immature behaviors that tend to be repeated in a routinely manner throughout the years. The permanent school staff that knows AR for years since he was 7 years-old (currently he is about 13 years-old), describe him as a young child that always confronted challenges in gaining friends among peers and tended to play with younger children at school. AR had difficulties in sustaining consistent conversations and congruent behavioral responses to a specific theme or a situation. Behaviors were "Childlish" even educators, teachers and therapists tried to mediate more mature behaviors and more congruent social and lingual responses.

On the physical level of organization, AR still find it difficult to organize his braille-books, papers and other items in his school-bag although he was trained and guided repetitively by different educators and occupational therapists across the last 6 years. In a similar manner, AR still has difficulties in constructional skills using various types and sizes of blocks, Cubes or Lego parts, although other congenitally-blind peers showed consistently-developing more complex organizational skills, sometimes astonishing, based on their

somato-sensory input, spatial skills, cognitive competencies, abstract ideational abilities and imagination as an outcome of a long-term social mediation and cognitive maturation enhances by school staff and parents of the young visually-impaired students.

Interview with AR's Parents

During the academic year 2015-2016 an interview was administered with AR's parents in order to gather information regarding his academic skills at home and his social interactions within the local community. His mother reported significant difficulties in teaching-learning process with AR at a daily routine. No minimal improvements were observed in his Braille tactual discrimination nor in phonological awareness although repeated trials were made intensively for teaching short and long phonemes in Arabic. In addition, no minimal progress was witnessed in basic Mathematical skills although different methods and strategies were applied at home using tactual materials and items in order to adapt concrete levels of cognitive strategies. Similar difficulties were reported in relation to the bilateral manual usage of Perkins-Braille in printing tasks at home. Bilateral coordination under the conditioning factors of precise timing and accurate sequencing of specific fingers in each printing trial was very unsuccessful, even frustrating and worthless. AR's father reported that his son presents difficulties in orientation and mobility even in a familiar environment of his own house and the nearby surrounding.

Discussion

Based on the various outcome measures described previously, both in formal academic domains and in the functional activities of daily living, suggest that AR presents a case of Gerstmann syndrome in the blind. Although the etiology of this syndrome was consistently reported as related to a lesion in the left parietal lobe, but in blind individuals it seems that the analogous area in the right hemisphere plays major roles in braille tactile discrimination and reading, motor-perceptual Braille printing skills, preliminary Arithmetic skills and Finger-gnosis.

One may argue that AR suffered already from right intra-ventricular hemorrhage of the newborn, Grade III in addition to the left intra-ventricular hemorrhage of the newborn, Grade IV (Hemorrhage in all ventricular space). In parallel to these facts, the author suggests that inserting the V/P shunt (supposedly the first, but more specifically the recurrent shunt insertion) through the superficial tissue of the Occipito-Parietal cortex and the Occipito-Temporal cortex evokes the diverse symptoms in AR, and intensifies the damage already exists in the intra-ventricular cavities. In other words, the damage across the neural organization in the surface of the cortical tissue, although limited to small surface, may have major impacts on functional abilities and even more diverse dysfunctions compared to those caused by subcortical areas. The insertion of the catheter through the right occipito-parieto-temporal junction in the cortical surface, and into the depth of its layers, is proposed to disconnect the neural networks in the specific point in this junction, thus evoking multiple dysfunctions related to the spatial perception and spatial orientation skills. It is important to argue that this cortical point of insertion does not have any task-specific nor skill-specific specialization, but rather a general "processor" for any spatial-related human function, both towards the human body itself (e.g., Motor planning, Spatial-motor coordination, Sequential performance, etc...) or towards the outer surroundings (e.g., Literacy, Reading and Writing, Arithmetic, Dressing, Turn-taking, Argumentation, etc...). An evidence for the necessity of cortical areas for reading in individuals with Alexia can be found in Johnson & Rayner (2007) were they proved that alexic patient (GJ) relied on cortical top-down processing while reading and had to activate decision making procedures that are characteristic of cortical centers when the bottom-up information was inaccessible due to subcortical lesions. This suggests that reading needs a complex neural networks that extends between cortical and subcortical areas and also between left and right Fusiform Gyri (Pyun, Sohn, Jung & Nam, 2008).

It seems that AR's neuro-anatomical lesions due to V/P shunt had significant direct implications on his pervasive functional limitations. Those lesions in the hierarchial layers of this associative area in the right posterior occipito-parieto-temporal junction seems to limit AR's ability to decode the spatial features of braille letters and words and the spatial demands of preliminary arithmetic skills. In addition, the disruption of mobility and orientation skills in familiar environments and the inability to achieve independence in performance of personal daily activities (particularly, dressing) seem to be related to the same preliminary disintegration of spatio-tactual

skills. Comparably, lesions in the surface of this right cortical area in sighted individuals could affect the visual word-reading skills, complex grapho-motor skills and handwriting skills. But, the compensatory advantage of vision and the visual attention in sighted individuals could, most probably, overcome the various limitations and enable functioning in these domains, at least within a satisfactory level. For instance, sighted individuals with Alexia succeed to read letter-by-letter in simple and slow sequencing, succeed to draw simple grapho-motor shapes and succeed to write simple and short words (Hillis, Newhart, Heidler, Barker, Hershkovitz & Degaonkar, 2005; Cohen, Henry, Dehaene, Martinaud, Lehericy, Lerner & Ferrieux, 2004; Lesniak, Soluch, Stepien, Czepiel & Seniow, 2014). Letter-by-letter reading, as a functional compensation in a case of Alexia, was proved to be based on the right analogous area of the left VWFA (i.e., R-VWFA) (Cohen, Martinaud, Lehericy, Samson, Obadia, Slachevsky & Dehaene, 2003). Other studies (e.g., Hamilton, Keenan, Catala & Pascual-Leone, 2000) suggested that also the striate and the pre-striate structures in both cortices play preliminary role in braille reading in early-blind individuals.

However, an individual with blindness, specifically with a congenital condition that prevented any visual acquisition of pre-literacy skills, may seek compensatory strategies from spatio-tactual resources in order to guide his spatial orientation, tactual-motor functions and adaptation to the diverse surroundings. In a case of an impairment in these neural resources, such as the case of AR, the blind individual may become deprived from alternative, compensatory resources. Consequently, every task and activity that is predominantly based on the integration of spatio-tactual and multi-sensory associative processing (e.g., braille reading, spatial orientation and dressing) becomes significantly limited, or even lost (more accurately, initially unlearned).

Based on the novel work of Siuda-Krzywicka, Jednorog, Marchewka & Amedi., *et al.* (2016) one can determine that the left visual word form area (VWFA) and the lateral occipital area (LOA) in sighted individual is responsible of visual reading. Does this associative cortical area play parallel role in tactual-spatial braille reading in blind individuals? Is the parallel cortical area in the right hemisphere play significant role in braille-related spatial discrimination, spatial-perceptual skills and orientation in the reading paper? Do blind individuals "visualize" the mental representations of the braille words they read? If this is the case, blind braille readers need an intact Posterior-Inferior Parieto-Temporal areas in the right cortex in order to acquire braille discrimination skills. Any lesions in this area, such as in a case of AR's V/P shunt and shunt revision, directly lead to a severe reading disability (Alexia of braille) and acquired pure Acalculia in braille.

Numerous empirical pedagogical and medical recommendations can be derived from the description of AR's case: (1) An intervention in occupational therapy should be planned and modified in order to establish alternative strategies for mobility and orientation and also for the basic activities of daily-living; (2) Alternative instruction methods and strategies should be comprehensively discussed, developed and applied in order to promote literacy skills only verbally due to Braille reading and printing inaccessibility; (3) Ophthalmological reassessments should be performed consistently in order to evaluate and estimate any pathological changed in the visual disk, optic nerve and the Retina, (4) Neuro-surgeons should take into consideration a clinical reasoning that focus on the anticipated future functional abilities and skills of the blind individual (e.g., braille reading, Arithmetic in braille and spatial-orientation) that are needed to acquire adaptive life-role and meaningful existence. Thus, a surgery for shunt implantation in blind infants should avoid the right posterior-inferior parieto-temporo-occipital cortical junction and associative areas in order to maintain spatial-perceptual and the spatial-tactual networks that are vital for blind individuals of all ages.

Novel developments in shunt design and manufacturing are continuously studied in order to create a "smart" shunt that can be controlled by digital devices or even smart phones. Such technological advancements can improve the prognosis of patients with shunt implantations and decrease the risks for shunt failures and complications. Nevertheless, all advancements do not compensate for the vital need to avoid the specific cortical associative areas that are pre-requisites for the blind individuals' future literacy and life-roles.

AR is currently assessed for health status, medical and physical conditions prior to a planned third V/P shunt surgery due to an increase in AR's height that requires an obligatory replacement of the existing catheter. Thus, alternative cortical areas should be considered for the planned surgery such as the left parieto-temporal and VWFA because it seem that it does not affect braille-related skill acquisition.

Risks, challenges and uncertain future will remain the companion of AR throughout his life. Therefore, Alternative methods of education, instructional strategies and rehabilitation interventions need shared efforts by both special-education teachers and paramedical therapists (e.g., Occupational therapists and O&M specialists) in order to create a new paradigm for intervention plans for similar cases. The new paradigm should be based on the strengths of AR and simultaneously avoid his impaired capabilities in a rehabilitative/compensatory approach of intervention both in academic tasks and in functional routines.

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