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HIDDEN VISUAL CAPABILITIES DIAGNOSED IN PRELINGUAL DEAF-BLIND

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Running head: Visual acuity, information processing.

Abstract

The visual acuity of twelve subjects, previously diagnosed as deaf-blind, were tested with a forched choice preferential looking procedure. Eight subjects possessed resolution acuity above the criterion for legally blind. Six of these had various degree of poor-to-normal eyesight. The four subjects with highest acuity were tested with the Fagan Test of Infant Intelligence which uses discrimination between known and novel faces in a preferential looking procedure to measure visual recognition memory. All four showed the ability to visual discrimination but various capacity for processing of visual information. Based on the fact that these subjects are previously diagnosed as deaf-blind with conventional methods, methods for assessing of visual acuity and visual processing in multi-handicapped persons are discussed.

Introduction

Often, when multi-handicapped persons come to vision assessment, they are passive, lack initiatives and gives few responses. Blind children are typically characterized by high degree of passitivity (Burlingham, 1975; Fraiberg; 1977), but passitivity may also be a behaviour consequence of, for example, lack of motor functions as in some people with cerebral palsy (von Tetzchner, 1987). Passitivity may also be caused by cognitive dysfunctions (Mundy & Sigman, 1989) or emotional problems. Informal observation procedures, clinical experience or optotype tests may fail to differential diagnose the cause of the passitivity, but only observe lack of responses to visual stimuli. The forced choice preferential looking (FPL) technique do not depend heavily on cognitive or motor responses and may therefore be a tool to indicate if reduced vision plays a part in the persons reduced response rate, or if the capability in the visual system is so high that the passitivity can not be explained from reduced vision.

Many reports indicate the usefulness of the FPL technique to assess visual acuity in multi-handicapped persons. In Mohn & Van Hof-Van Duins (1983) study of children with neurological disorders, PL acuity were obtained for 23 out of 32 patients tested. FPL and pattern VEP were used because accurate assessment of visual functions with conventional, clinical methods is difficult or impossible in children with neurological disorders, particularly when the disorder is accompanied by more or less severe psychomotor retardation (Mohn & Van Hof-Van Duin 1983). Mohn and Van Hof-Van Duin (1983) used an operant PL technique for children of 1,5 years or older. They also practiced, as a technique, to turn the child slowly to and from between grating and the blank. Very often, as they describe, the childs gaze would then "stick" to the grating. This technique was used to avoid the stimuli from falling in a blind erea of the child's visual field and to overcome possible oculomotor problems. The technique also facilitated observations when a strong nystagmus and/or a strabismus were present. Mayer, Fulton & Sossen (1983) studied pediatric patients with developmental disorders and estimated PL acuity in 79% of the patients on the 1st attempt. They also found that many children who were "visually inattentive" despite the absent of major ophthalmological abnormalities, were testable with PL procedures, but had poor acuity. Birch & Bane (1991) used FPL successfully in 132 children with cortically visually impairment. From results they suggest that FPL estimates provide information about the cortically, visually impaired childs ability to use vision in every day life, and may therefor be useful for evaluating treatment response and establishing guidelines for educational materials for these infants and children. O'Dell, Harshaw & Boothe (1993) concluded that the acuity card procedure is a useful vision screening tool for individuals with sever or profound mental retardation. 271 individuals with sever and profound mental retardation were tested with FPL procedure, and acuity values were evaluated in terms of test-retest as well as inter-, intra- observer reliability. Lennerstrand, Axelsson & Anderson (1983) examined visual acuity successfully in 21 of 26 mentally retarded children and young adults with operant preferential looking. They found a good correlation between diagnosed ocular abnormalities and the visual acuity results. Hall, Orel-Bixler & Hagstrom-Portnoy (1991) included FPL as a useful technique to assess vision capabilities of multi-handicapped persons. Hertz & Rosenberg (1992) tested 77 spastic children with mental retardation, and found a very good test-retest reliability. They concluded that their results show a high success rate for the use of acuity cards to evaluate the visual acuity of severely disabled, mentally retarded children.

However, there are reports that indicates limitation when FPL is used in visual assessment suggesting that special precursors should be implemented in the testing procedure. The behavioural acuity of infants with foveal abnormalities has been found to be only slightly lower than that of normal infants, and often within the normal range (Mayer, Fulton & Hansen, 1985), suggesting the possibility to overlook persons which lack central vision when assessing visual acuity with grating FPL.

A grating-FPL do only provide a pure measure of resolution acuity without involvement of high-level vision (Van Hof-Van Duin, 1989: Mohn & Van Hof-Van Duin, 1991). Results from a FPL grating test will therefore not give a hint to how a person tested will solve visual tasks which involve recognition, identification and visual memory. Another category visual acuity tests are the recognition tests (optotype tests). These tests involves cognitive functions as recognition and identification, and thereby testing functions in areas of the brain such as the parietal and temporal cortex (Van Hof-Van Duin, 1989; Mohn & Van Hof-Van Duin, 1991). However, recognition tests do not differ between resolution acuity and high-level vision, and will therefor not differentiate between visual acuity and cognitive functions like recognition and memory as cause when the response rate is low in multi-handicapped persons. One way to solve this problem may be to use a FPL grating test for visual acuity, and a cognitive test for processing of visual information from primary visual cortex to other parts of the brain. However, cognitive tests for multi-handicapped persons are hard to find, because most cognitive tests involves verbal instructions and complex responses. A relative new test that may be of interest for the purpose of dividing between visual acuity and processing of visual information in multi-handicapped persons is the Fagan Test of Infant Intelligence (FTII) (Tetzchner, Jacobsen, Smith, Skjeldal, Heiberg & Fagan in press). The FTII is a paired comparison test of visual recognition memory (Fagan & Detterman, 1992). The test is essential self instructive: consisting only in directing the subjects attention towards a screen displaying two pictures. Following exposure to one picture, the previously exposed picture and another picture are briefly paired. Normal infants and children tend to devote more attention to the novel picture. A version of the FTII for ages above two years has recently been developed, and was used in the current study. The new version makes comparison with normal functioning children and youth possible. The novelty score derived from the Fagan test does not have an age equivalent. Performance on the test may still provide indirect support for better cognitive abilities in ereas that depend on memory and perception than in ereas related to planning and response. However, the main purpose of the study was to gain insight to the subjects abilities to remember and process visually presented material, and to use two tests that assessed visual acuity and processing of visual information independently. The measurement principle (differential fixation to novel over the previously seen facial pattern) in the FTII is very much like a habituation procedure, and indicates that very simple habituation procedure can be applied to the group of multi-handicapped persons who do not respond on FTII for reasons like problems with sustained attention or severely reduced vision, to decide whether visual information is processed from visual cortex to other ereas of the brain or not. Example on such a simple procedure may be presentation of a filled square randomly to the right or to left in the visual field in a series of five presentations. If the inspection time is systematically reduced for each presentation, it is an indication of recognition.

Combining a FPL grating test for vision and visual processing test for multi-handicapped persons may both give a reliable capacity of pure resolution acuity and visual processing.

The present study applies the FPL technique, using Teller Acuity Cards (TAC) to assess visual acuity in multi-handicapped, mentally retarded people diagnosed as prelingual deaf-blind. Registration and diagnoses of prelingual deaf-blind persons in Norway have mainly been based on informal observation procedures and clinical experience. In a few cases optotype tests have been in use. For some of the subjects in the study, The FTII is also applied.

Method

In Norway, the national health department has a constant ongoing registration of deaf-blind persons, and at the present time there are registered 86 prelingual deaf-blind persons. Twelve mentally retarded children and youths diagnosed as prelingual deaf-blind participated in this study. The sample comprised 15% of the born deaf-blind population in Norway. Deaf-blind children were selected to this study because they were considered particularly difficult to test, and because they were already diagnosed from informal observation or conventional methods that do not differ between visual acuity and processing of visual information. The 12 participants in this study constituted the born deaf-blind population living in or near by Oslo.

Visual acuity was assessed with the well known Teller Acuity Cards (TAC). The forced choice preferential looking, blind testing and staircase procedures were followed. Constraints and limitations with the method are also taken into consideration when the results are presented. With a flexible test procedure (within the demands for the reliability for the test) eleven of our twelve subjects could be tested in a single session lasting 20 to 80 minutes. For some of the subjects adjustments in the testing procedures were necessary to accomplish the test. Eight of the participants were tested either in their home or at their school. Three had to be tested laying down. Under both of this two conditions the luminance was carefully measured. When subjects had to be tested laying down, they were laid head towards the window, resting their head on a pillow, so that daylight luminated the cards. Seven subjects did not show the sustained attention to complete the test in one trial. Restarting the test, each trial started with the card with lowest spatial frequency, then the card a half octave below the last card responded to in the previous trial was presented. All participants were retested except for subject 7 and 8 who moved to another part of the country shortly after the first testing. No retesting took place less than twenty days after the first testing.

Four out of the 12 subjects were also tested with the video edition of the child and adult version of the Fagan Test of Infant Intelligence for age 2 years and above (see description in Fagan and Vasen in press). Six subjects were considered to have visual acuity required for FTII (6,5 cy/deg or better), but the two subjects with acuity 6,5 cy/deg moved to another part of the country before the FTII were carried out, so, the four subjects tested have acuity 9,8 cy/deg. or better. The subjects received 20 novelty problems, all problems involving comparison of face images presented on a video screen. Each of the 10 first problems consisted of a 5-seconds familiarization period to one target, then pairing the briefly studied pattern with left-right position reversed from one period to another (immediate memory). Following this, there were 10 problems where a new picture was presented together with a previously exposed target for two 2-seconds pairing (delayed memory). Visual recognition was measured by differential fixation to the novel over the previously seen facial pattern. At each session different targets were used, but inspection time and test time were the same at the two occasions. The observer, hidden behind a piece of cardboard behind the video screen, could see the corneal reflections on the targets through a 0.64 centimeters hole in the cardboard. The observer pressed one of the two mouse buttons of the computer according to which side the child was looking. Number of periods the child was looking towards each target (fixations) was also registered. Average inter observer reliability in the measurement of differential fixation is 0.94 (Fagan and Detterman 1992)

Results

PL acuity were obtained in 11 of the 12 participants. Test and retest results are shown in table 1. Subject details (age, diagnosis and refraction) are also presented in table 1. The subjects were assign to numbers after visus. Three subjects showed pure acuity (2-4). Eight subjects possessed acuity above the criterion for legally blind (5-12). The difference between test and retest for each individual did newer vary more than a half octave.

Insert table 1 about here

Preference for new faces for the four persons with acuity better or like 9,8 cy/deg are shown in table 2. One person (11) showed normal processing speed. Subject 8 and twelve showed moderate to slow processing speed, whereas subject 10 possessed the ability to visual discrimination, but a very slow processing speed.

Insert table 2 about here

Discussion

With a suitable adapted FPL procedure visual acuity could be determined in 11 of the 12 deaf-"blind" children; eight subjects scored well above the criterion for legally blind (> 4,8 cy/deg is by convention approximately 0,2 Snellen equivalent, as far as visual information processed by the eye and the retina and the primary visual cortex is concerned), and six out of these proved to possess various degree of poor-to-normal eyesight. Of the four persons tested with FTII (also PL technique) one showed normal speed of processing information, two showed moderate to slow, and one showed very slow processing speed.

Previously diagnosing practice by conventional methods like observations and recognition acuity tests overlooked both the visual capability in eight subjects and processing capability in four subjects. The different outcome of earlier diagnosing and this study suggests that assessment in multi-handicapped persons ought to be carried out with test procedures that are more or less free from instructions and do not depend on complex and conscious responses, like the PL procedure used in this study. The result in the current study also suggest that assessment of visual acuity and processing of visual information from visual cortex to other parts of the brain should be measured independently in multi-handicapped persons.

For the four subjects that had visual acuity above the criterion for legally blind, but were not tested with FTII

(Person nr. 5,6,7 and 8. 5 and 6 were considered to have to reduced vision for reliable testing with the FTII, whereas 7 and 8 were considered to have vision required for face recognition, but moved to another part of Norway before retest with TAC and the testing with FTII took part), behavioural states were observed. They all showed periods of wakefulness that are considered necessary to process information from stimuli, which indicate that visual processing also occur (Wolff, 1987). For person 5 and 6 a simple habituation procedure were carried out. For one of them results indicated that visual processing occurred, for the other we did not manage to complete the procedure. However, data from the observation of behavioural states and the habituation procedure are to preliminary to be presented, but are examples of simple methods that may give an impression if visual discrimination and visual processing occur, and may together with a resolution acuity test complete an assessment procedure for visual acuity and visual processing to brain ereas concerned with memory, identification and recognition.

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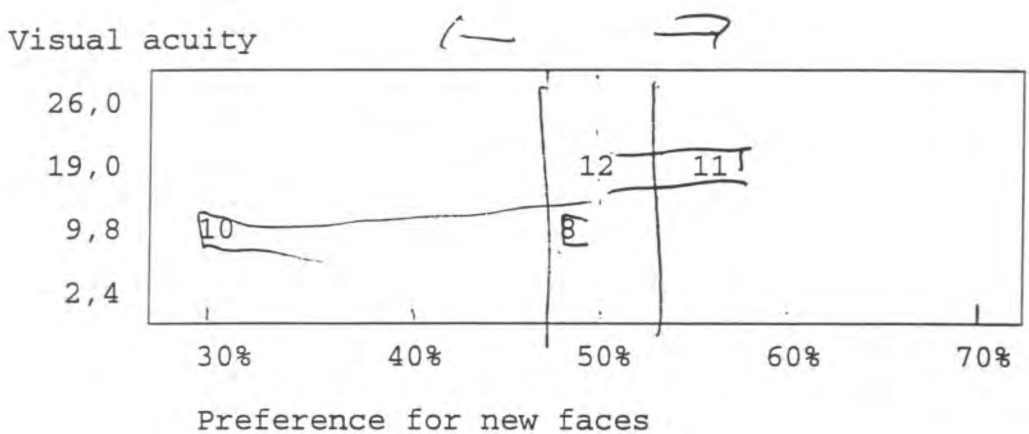
Table 1. Subject details and visual acuities measured by the acuity card procedure. All subjects were tested with correction for refractive errors. NR in the result sections stands for not retested. Numbers are assign after visus.

Table 2. Relation between visual acuity and preference for new faces for subjects with acuity 9,8 cy/deg or better. The numbers in the table refer to the subjects numbers in table 1.

Table 1

Subject details				Results	
Subjects	Age	Diagnosis	Refraction	Cy/deg test	retest
1	18	Cataract	-	-	-
2	23	Cataract	-	1,6	2,4
3	18	Macula Degen.	+1,0; Bino	2,4	2,4
4	30	Cataract	+3,5; Bino	2,4	1,6
5	4	Cataract Glaucom	+3,0;+3,5 Astigmatis	4,8	4,8
6	19	Ceratoc.R.E.	-4,5; L.E.	4,8	4,8
7	27	Cataract	+2,0; R.E	6,5	NR
8	23	Cataract.R.E. Glaucom.L.E	-	6,5	NR
9	28	-	Astigmatis	9,8	9,8
10	17	-	+5,0;+4,0	9,8	9,8
11	20	Catara.L.E	-2,0	19,0	13,0
12	14	Catara.R.E	-2,0	19,0	19,0

Table 2


Visual acuity
26,0
19,0
9,8
2,4
12
11
10
8
30%
40%
50%
60%
70%
Preference for new faces

LETTER TO THE EDITORS

HIDDEN VISUAL CAPABILITIES IN MENTALLY RETARDED SUBJECTS PREVIOUSLY DIAGNOSED AS DEAF-BLIND

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Abstract - The visual acuity of twelve multi-handicapped, mentally retarded subjects, previously diagnosed as deaf-blind, was measured on two occasions with the Teller Acuity Card (TAC) procedure. Eight subjects scored above the criterion for legally blind and the results of six of these indicated various degree of poor to almost-normal eyesight. To evaluate high-level vision four subjects were tested with the Fagan Test, testing visual recognition memory for faces in habituation variant of the preferential looking technique. The results for three subjects showed evidence for perceptual recoggnition. It is concluded that TAC combined with the Fagan Test may detect perceptual capacities unnoticed by clinical observation.

A number of reports indicate that the forced-choice preferential-looking technique (FCPL), originally developed for testing vision in infants (Teller, 1979; Mohn & Van Hof-van Duin, 1991), may be successfully applied to measure visual acuity in multi-handicapped children and adults who are not testable with conventional clinical ophthalmological procedures (Mohn & Van Hof-van Duin, 1983; Hall, Orel-Bixler & Haegstrom-Portnoy, 1991; Harshaw & Boothe, 1993). The FCPL is based on the infant's tendency to fixate any visible object in an otherwise empty field (Fantz, 1958), and does not require perceptual recognition, language skills or coordinated manual responses. When measuring visual acuity with FCPL, black-and-white luminace gratings whose mean luminance matches a neutral gray surround, are presented randomly to the left or right, and an observer who is blind to the grating presentation, watches the subject's eyemovements and guesses the location of the grating; visual acity is the highest spatial frequency, in terms of number of cycles per degree visual angle (c/deg), where the observer guesses the location of the grating with a probability of .75 correct (Teller, 1979).

Mohn and Van Hof-Van Duin (1983) used the preferential-looking technique to test children with neurological disorders, and were able to determine visual acuity for 23 out of 32 subjects tested despite severe psychomotor retardation in some of the subjects. For children of 1,5 years or older preferential looking was paired with an operant procedure where subjects were rewarded for a correct response. To avoid the stimuli from falling in a blind area of the child's visual field and to bypass oculomotor problems, they also practiced to turn the child slowly back and forth between grating and blank, and observed that the child's gaze would then "stick" to the grating. This procedure was also useful when testing children with strong nystagmus and/or strabismus (Mohn & van Hof-van Duin, 1983). Lennerstrand, Axelsson and Anderson (1983) likewise used an operant preferential looking procedure to measure visual acuity in mentally retarded children and young adults, and obtained an acuity estimate in 21 of 26 subject tested. They also found high correspondence between diagnosed ocular abnormalities and the visual acuity estimates. In another early study Mayer, Fulton and Sossen (1983) tested a large sample of pediatric patients with developmental disorders and determined preferential looking acuity in 79% of the patients on the first testing session. They further observed that many children who were "visually inattentive" despite the absence of major ophthalmological abnormalities, were testable with preferential looking procedures. Birch and Bane (1991) used FCPL to test 132 children with cortical visual impairment. They conclude that the FCPL acuity provides valid information about the child's ability to use vision in every-day life, and may be useful for evaluating treatment and establishing guidelines for educational materials for these children.

Most of these studies were laboratory based with different types of apparatus and test conditions. However, Teller and her associates have develped standard test situation for clinical use, consisting of a set of calibrated photographic cards and a carboard stage (Teller, McDonald, Preston, Sebris & Dobson, 1986). Recently O'Dell, Harshaw and Boothe (1993) tested 271 individuals with severe mental retardation with the Teller Acuity Cards (TAC). They analysed test-retest as well as intra- and inter-observer reliability, and concluded that the acuity card procedure is a useful vision screening tool for these patients. Similar conclusions were reached by Hertz and Rosenberg (1992) who tested 77 spastic children with mental retardation.

These studies show that the varieties of the FCPL including the technically simple acuity card procedure, are valid and reliable techniques for determining visual acuity in severely retarded and disabled persons. In the present study we have used the TAC to test a selected group of subjects, namely multi-handicapped, severely mentally retarded persons that were already diagnosed as deaf-blind.

A reliable FCPL acuity obtained in severly disabled persons does not by itself tell us anything about her/his visual perception in the conventional sense of visual recognition. It is conceivable that FCPL performance some subjects may be goverened by subcortical mechanisms of vision which are not involved in conscious perception. Studies of "blind-sight" in humans with unilateral lesions of the visual cortex (Weiskrantz, 1986; Ruddock, 1991) show above chance visual performance on detection and localization without conscious awareness of the stimuli, and in some of these cases the performance must be controlled by subcortical mechanisms (e.g. Magnussen & Mathiesen, 1989). In this sense FCPL only index the resolution of the sensory system, and do not tell us about high-level vision (Van Hof-van Duin, 1989; Mohn & Van Hof-Van Duin, 1991). To conclude that the person has functional vision some additional indication of pattern or object discrimination is required. In the present paper a subsample of the subjects was tested with the Fagan Test which is a preferential looking test of visual recognition memory for faces (Fagan & Detterman, 1992).

Method

Subjects

The National Health Department keeps a continous record of deaf-blind persons born and living in Norway; in 1994 a total of 86 deaf-blind persons were registered of a population of about four million people. In the present study 12 mentally retarded children and young adults from that register were tested. The sample comprised 14% of the born deaf-blind population in Norway, and all the born deaf-blind living in the Oslo region where the study was conducted. Six of the participants were females and six were males, varying from four to 30 years of age (Table 1). Using the criteria of DSM-IV (American Psyciatric Association, 1994) two subjects were diagonosed as moderately mentally retarded (subjects 2 and 7), five as severely retarded (subjects 1, 4, 8, 10, 11) and five as profoundly retarded (3, 5, 6, 9, 12). All subjects were muli-handicapped in the sense that they had combined sensory impairments; in addition, the subjects diagnosed as profoundly retarded exhibited severe psychmotor retardation. All subjects were pre-linguistic in the sense that they were unable to communicate in any formal language, hand-language or otherwise.

In Norway assessment and diagnoses of the visual capacities of the mentally retarded deaf-blind persons are based on a combination of ophthalmological examination, careful clinical observation and informal testing of attention and responsiveness to visual stimuli, carried out by trained personell. Ophthalmological data were available from the medical records of all the subjects in the present sample, and are summarized in Table 1. Nine subjects had referactive errors and these subjects were fitted with lenses prior to testing, except for one subject (No. 4) who refused to wear spectacles.

Procedure

Visual acuity was measured with the Teller Acuity Cards, using the forced choice procedure, blind testing and a psychophysical staircase procedure as recommended. The TAC consists of 15 neutral gray cards containing a 12,5 x 12,5 cm square-wave grating at a distance of 7 cm from a tiny peephole in the center; grating contrast is 0.5 and background luminance matches the space average luminance of the grating. The spatial frequency varies between 0.32 and 38.0 c/deg for a viewing distance of 55 cm. The acuity cards are presented in a rectangular window in a larger cardboard screen shielding the subject's visual field.

Eight of the subjects were tested in either home or institutional settings, four subjects were tested in a standard laboratory setup. The subjects were either sitting in a supporting chair or, when not possible, were tested lying down resting on a pillow. In these (three) cases the cardboard screen had to be abandoned, but distance markers and lighting conditions were improvised so that in all cases test distance and card luminance conformed to test requirements. Testing was binocular.

When the subject was considered attentive by the tester, a card with a grating randomly to the right or left was presented. The tester, who did not know the location of the grating, observed the subject's eye movements through the peephole in the centre of the card and guessed the location of the grating. Presentation started with a low spatial frequency card and proceeded in half octave steps towards higher spatial frequencies: when the observer's guess was correct a higher spatial frequency was chosen on the next trial, when the observer's guess was wrong, a lower spatial frequency was chosen. In the present study acuity is the lowest spatial frequency of four-to-six reversals of the stair-case on consectitive trials. In several cases it was not possible to catch the subject's attention over a sufficiently long time to cmplete testing in a single run. In this case the test was run a second time after a suitable break, starting once again with the lowest spatial frequency card and then proceeding directly to the spatial frequency half an octave below the last card producing an correct guess by the observer on the previous run. With a flexible procedure and suitable improvisations visual acuity could be determined for 11 subjects in a single session lasting from 20 to 80 minutes; one subject (No. 5) showed no response to any test card in either test or retest sessions.

Except for two subjects who moved to another part of the country shortly after the first test session, subjects were tested on two occasions with a minumum interval of twenty days between test and retest. All tests were performed by an experienced observer (author KJ).

Insert table 1 about here

Results and discussion

Visual acuity estimates were obtained in 11 subjects, one subject (No 5) with binocular cataract did not respond to any test card during in either test or retest sessions. The results are shown in Table 1, with subjects assigned numbers according to age, for test and retest sessions independently. Values are given in spatial frequency (c/deg) and in Snellen equivalents. The test-retest reliability is very good with acuity estimates differing by less than half an octave, confirming previous studies (Hertz & Rosenberg, 1992; O'Dell *et al.*, 1992)

For three subjects with ophthalmological diagnoses macula degeneration and binocular cataract (No. 4, 9 and 12) blindness was confirmed in the sense that the visual acuities measured by TAC corresponded to the legal definition of blindness proposed by WHO (spatial frequency of 2.4 c/deg, Snellen acuity of 0.1). The remaining eight subjects all exhibited visual acuities above this criterion, and in two the subjects (No. 2 and 7) TAC acuities approached normal vision; the reminding subjects exhibiting various degree of poor to moderate visual acuity. To get an idea of the visual performance of these subjects, consider that a visual acuity of 6-8 c/deg corresponds to the acuities measured in the six month old baby (Mohn & Van Hof-Van Duin, 1991), the rod monochromat (e.g. Greenlee, Magnussen & Nordby, 1988) or the adult domestic cat (Crawford, Anderson, Blake, Jacobs & Neumeyer, 1990). Thus, provided vision is functional, these patients are by no means blind.

However, as noted above, FCPL grating acuity measures do not index high-level vision in infants or disabeled persons (Van Hof-Van Duin, 1989; Mohn & Van Hof-Van Duin, 1991). They do not tell us about perception proper, how the subject will perform on normal visual tasks involving recognition and identification. Conventional recognition tests (optotype tests) on the other hand, which do involve these higher-level perceptual-cognitive functions, confound problems of visual resolution, high-level vision and cognitive-communicative skills when applied to such persons. Therefore, when deficits in perception are suspected a preferential-looking test should be supplemented with a perceptual test measuring visual recognition and vice versa. However, most tests of recognition require that subjects understand and can follow verbal instructions and make an appropriate response. They are therefore not applicable to the current subjects.

To evaluate perceptual recognition we decided to test those subjects who had a sufficiently high acuity score with the Fagan Test (Fagan & Detterman, 1992), recently applied to evaluate high-level vision in Rett's syndrome (von Tetzchner, Jacobsen, Smith, Skjeldal, Heiberg & Fagan, 1996). The Fagan Test is a test of visual recognition memory for faces based on a habituation variant of the preferential looking procedure: On a given trial a picture of a face is first presented for inspection, and then paired with another face, and the subject's preference is measured by looking time. If the subject systematically favors either the familiar or the novel face, perceptual discrimination must be present.

FACE DISCRIMINATION

Method

Face discrimination was tested by the computer edition of the child and adult version of the Fagan Test for age two years and above (Fagan and Vasen, in press). Six of the subjects in the first study were judged to have sufficiently high preferential looking acuity to be tested with the facial pattern (6,5 c/deg or better, subjects No.

2, 3, 7, 8, 10, 11) but two of these (No. 8 and 10) moved to another part of the country before the Fagan Test was available, leaving four subjects with acuities of 9,8 c/deg or better.

The Fagan Test consists of 20 novelty tasks, all involving comparison of achromatic pictures of human faces presented on a computer screen. There are two blocks of 10 trials, one testing immediate memory and one testing delayed memory. In the immediate memory condition each trial consists of a 5-sec presentation of a single face, immediately followed by a 4-sec test presentation of the "familiar" face paired with a novel face which switch left-right position after two seconds. In the delayed memory condition, the novel face is presented together with a face shown on one of the previous trials. The computer screen subtended 30 deg visual angle, the face stimuli subtended 13 deg visual angle, and they were separeted by a 4-deg blank space.

Visual recognition is measured by the differences in looking time between the novel and the familiar face. The observer, hidden behind a cardboard screen observes the corneal reflections of the targets through a tiny hole, and records left or right looking by pressing one of two mouse buttons. Testing was performed by a well-trained observer (author LS). Average inter-observer reliability of trained observers in recording differential fixation is 0.94 (Fagan and Detterman, 1992).

Results and discussion

Right column in Table 1 show preference scores on the Fagan test in terms of accumulated looking time for new faces in per cent of total looking time. The standarization of this test defines 47/53 per cent criterion for displayed preference (Fagan, Singer, Montie & Shepard, 1986). Three of the four subjects tested scored byond this limit, with two subjects (No. 3 and 7) exhibiting a strong preference for either the familiar or novel face (33.3 and 57.0 % looking time), one subject (No. 2) showed moderate preference for the novel face (53.7 %), and one subject (No. 8) showed no preference in either direction according to the test criteria (48.9 %).

The results indicate that three of the subjects possess the perceptual-cognitive skills necessary to discriminate and remember human faces, whereas for one person such skills have not been demonstrated. Note, however, that the logic of the test only permits affirmative conclusions. Strictly speaking, chance performance on the Fagan test does not imply that perceptual discrimination is not present, only that it does not turn up in a systematic across-trials perceptual preference pattern. However, the absence of face preference is an indication that the subject do not possess functional vision despite a moderate visual acuity score on the TAC. Table 1 further suggest that it is not visual acuity that sets a limit to face discrimination.

GENERAL DISCUSSION

The results of the present study show that with suitable testing procedures it is possible to obtain evidence for perceptual capacities that are otherwise bypassed by clinical observation. With the TAC procedure visual acuity could be determined in 11 of 12 severely mentally retarded children and young adults that had been diagnosed as deaf-blind by the medical examination routines of the Norwegian halth care system for multihandicapped people, and eight of these proved to possess various degree of poor to almost-normal normal visual acuity. While preferential looking acuity does not by itself prove the presence of perceptual processing, tests of visual recognition carried out on four of the subjects gave clear evidence for perceptual recognition in two subjects, and we might suspect similar capacities in some of the subjects that were not tested.

Why were perceptual capacities not previously detected in these subjects? The main factor of course is that these persons are very difficult to test by any type of test, and since multihandicapped subjects frequently are passive, lack initiative and do not respond to visual stimuli, their behaviour might be misinterpreted as a sign of blindness. Passivity is typical for blind children (Burlingham, 1975; Fraiberg, 1977), but may also be a consequence of, for example, disturbed motor functions as in some people with cerebral palsy, cognitive dysfunctions (Mundy & Sigman, 1989) or even emotional disturbances (Beck, Stanley, Averill, Baldwin & Deagle, 1992; Wells & Mattews, 1994) . More informal observation procedures and clinical judgment may fail to identify the cause of passivity. Forced-choice preferential looking techniques which do not rely heavily on higher-level cognitive processes or motor responses would seem to be an ideal tool evaluating the sensory capabilities of these patients. Combining the TAC with some measure of visual discrimination, such as the Fagan Test, we can further decide if the visual communication channel is open or closed. In order to develop efficient habilitation programs for multi-handicapped persons it is necessary to be able to differentiate between sensory deficits and other factors limiting interaction and behavioural performance. TAC and the Fagan Test should both be useful tools in this process.

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Table 1

Subject details					Visual acuity	
Sub.	Sex	Age	Diagnosis	Refraction	C/deg Test-ret.	Snellen Equiv.
1	F	4	Cataract Glaucom	+3,0;+3,5 Astgmatism	4,8- 4,8	0,2
2	F	14	Cataract.R.E.	-2,0;R.E.	19,0-19,0	0,7
3	M	17	-	+5,0;+4,0	9,8- 9,8	0,4
4	M	18	Macula Deg.	+1,0;Bino	2,4- 2,4	0,1
5	M	18	Cataract	-	0- 0	0
6	F	19	Ceratoc.R.E.	-4,5;L.E.	4,8- 4,8	0,2
7	F	20	Catara.L.E.	-2,0;R.E.	9,0-13,0	0,5
8	F	23	Catara.R.E. Glaucom.L.E.	-	6,5- NR	0,35
9	M	23	Cataract	-	1,6- 2,4	0,1
10	M	27	Cataract	+2,0;R.E.	6,5- NR	0,35
11	F	28	-	Astigmatism	9,8- 9,8	0,4
12	F	30	Cataract	+3,5;Bino	2,4- 1,6	0,1

Table 2

Subject	Fagan	Visual Acuity
2	53,7%	19,0
3	33,3%	9,8
7	57,0%	13,0
8	48,9%	9,8

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17.09.96

Also transmitted via E-mail 6.09.96

Dear Prof. Zrenner,

I am writing on behalf of the authors (S. Jacobsen, S. Magnussen & L. Smith) of MS VRE2214 "Hidden visual capabilities in mentally retarded subjects diagnosed as deaf-blind" rejected for publication as a Letter to the Editors in the Clinical Vision Sciences section. We received two reviews, one very favourable stating that the study is a useful one, and the "The paper is well written and the author's findings are clearly reasoned". The second is extremely negative, pointing to major difficulties relating to "sample size, characterization of subjects, overestimation of acuity and onserbver bias".

I appreciate the difficulties sometimes facing an editor ,and realize that it is not considered "comme il faut" to argue with reviewers, but the negative reviewer displays such obvious lack of understanding of the issues addressed in this paper, in addition to superficial and biased reading of the MS , that it is difficult to take his/her comments completely seriously. I'll address the reviewer's points as they were numbered in the report:

1. "The number of subjects is too small for useful generalization to other populations..." Populations of what? Mentally retarded? Deaf-blind? Generalizations to whom? This is not an attempt to establish norms of how well mentally retarded or deaf-blind persons see. This is a targeted sample, comprising some the most deeply mentally retarded persons living and selected precisely because they had already for a various number of years been diagnosed, and treated, as deaf-blind by one of the most advanced public health-care systems in the world. The point is a methodological one, namely that because of gross brain disturbances and motor disturbances, it could be that in some of these cases, preferential looking might detect som vision and a test of recognition might reveal basic object discrimination. Actually, a single case would be sufficient to make the point. The prevalence of this diagnosis is 1 pr 40.000 (twice as frequent as e.g. rod monocromats), and we were able to test 12. The reviewer's objection therefore misses the target, sample size is not the question here.

2. Regarding the subject description the reviwers write " Is this just a terminology usage , where "deaf-blind" does not necessarily mean either deaf or blind?" Is this supposed to be a joke? Does the reviewer believe that a serious health-care system diagnoses and subsequently treats people as deaf-blind when they are not believed to be? It is also stated in the MS that the diagnosis was based on behavioural judgement from trained personell including neurologists. Blindness in this case is based on visual behaviour (with the more informal methods available, before TAC and Fagan). Since these are brain injured subjects, it is of course likely that blindness in many cases were ascribed to cortical impairment. But this should be obvious from the type of patients we have tested.

3-4. With this type of patients one has too balance the optimal solution against what is practical. Our stair-case would not converge on 50% correct, because we did not calculate the mean of the reversals, but used the lowest spatial frequency involved in four-to-six consecutive reverals, that is the frequency visible each time. If anything, this is a conservative estimate. This is described in the MS, but obviously gone undetected by the reviewer.

5. "The time required for testing each subject suggests that the procedure used was not an acuity card procedure. Testing should be completed in 10-15 minutes". Yes, in normal alert babies. Do the reviewer believe we are testing Helen Kellers? Some of these patients are close to just vegetate, they cannot even crawl, they have no motor control, they doze off or get fits. Testing can only be performed when the subject appears a little alert and with eyes in the correct direction. However, with patience and suitable adaptation of TAC is is possible to obtain data from at least some such patients, who may not be completely without visual perception. It is quite possible that strict adherence to a formal procedure is the reason for somewhat lower reliability in other studies.

6.-8. The concept test-retest reliablity refers to tests conducted on two different occasions, inter-tester reliability refers to the case when two (or more) observers score the same test-protocol or observe simultaneously. If both time and observer change, one cannot distangle the two sources of variability . The reviewer's implicit recommendation that different observers should be used in test and retest is therefore a bad one. In particular in this case where we might also expect changes in state of the patients.

10. This point has been answered above - the estimation is probably on the concervative side.

12. 6.5 c/deg acuity was chosen for selection to the Fagan test so that interpretation of negative results was not confounded (i.e. due to low visual acuity or lack of perceptual recognition). Most previous previous reports give infant acuity of about 6 c/deg around six months.

In conclusion, I really do not see the basis for this reviewer's negative evaluation. This letter describes a clinical study, attempting to apply preferential looking tests of both detection and (for the first time) recognition to the small group of deeply mentally retarded subjects which in addition are believed to be deaf-blind . Our surprising finding strongly suggest such tests should be established as clinical practice. And I believe that it deserves a letter in a clinical vision section.

I would much appreciate if you would reconsider you decision in the light of the above comments.

Yours sincerely

Svein Magnussen

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02.08.96 08:33/Prof.ZR/rt
Re: Ms No. VRE2214

Dear Dr. Jacobsen,

Thank you very much for submitting your manuscript "Hidden visual capabilities in mentally retarted subjects diagnosed as deaf-blind" (VRE2214) to VISION RESEARCH, Section 5: CLINICAL VISION SCIENCES.

Unfortunately, as you can see from the reviews enclosed, there seem to be major problems with your paper and I regret having to inform you that the paper cannot be accepted for publication in its present form. However, I hope that the critical comments of the reviewers will prove of some value for you and your future scientific work.

I am very sorry that I have to decline publication but please understand that I have to follow the reviewer's recommendations. Nevertheless, I would hope you will consider our journal again for the publication of some future study.

With best regards

[redacted]
Section Editor of VISION RESEARCH
Section 5: CLINICAL VISION SCIENCES

enclosure:
referees' reports

#2

Re: Ms. VRE 2214 Hidden visual capabilities in mentally retarded subjects diagnosed as deaf-blind

by K. Jacobsen, S. Magnussen, L. Smith

This paper is concerned with spared visual capacities in children or young adults diagnosed as deaf-blind. The procedure that the authors used was developed to assess these abilities as well as recognition memory in a covert, implicit manner, thereby avoiding tests that are dependent upon complicated instructions or effortful processing. The results clearly support the usefulness of such procedures in the assessment of visual functions, at least in a subgroup of mentally retarded subjects. The paper is well written and the authors' findings are clearly reasoned. There are, however, several minor issues which should be addressed.

1. The introduction is very long relative to the general discussion, and the description of individual studies (on pages 3 and 4) could be shortened considerably.

2. It would be useful for the reader to have more detail about the general cognitive performance level of the two subgroups who were diagnosed as severely or profoundly mentally retarded according to DSM-IV.

3. Results for the Face Recognition task. In the Methods section, the authors mention that immediate and delayed recognition are assessed separately. Do the results presented in Table 1 refer to an average of both subtests? It would be informative to have some data on the findings for both delays.

4. It is interesting to note that 3 of the subjects who were able to signal simple needs (2,7,11) were the same subjects who performed relatively well on many visual tests. For example, subjects 2 and 7 showed TAC acuities approaching normal vision and subject 11 also did better than most of the

other subjects. In addition, subjects 2 and 7 showed some degree of face recognition. The authors might want discuss whether severity of mental impairment may relate to performance on the perceptual tasks.

5. In the introduction, the authors discuss the possibility that FCPL performance may at least partly be mediated by subcortical mechanisms. They might readdress this issue in the General Discussion; i.e. they should discuss whether their findings add new evidence for this hypothesis.

#1

Review of VRE 2214, "Hidden Visual Capabilities in Mentally Retarded Subjects" Diagnosed as Deaf-Blind" by Jacobsen, Magnussen, & Smith.
p. 1

This manuscript reports a study of grating acuity in 12 multiply handicapped individuals. Major difficulties with the study and report include sample size, characterization of the subjects, and problems with methodology leading to possible overestimation of acuity and observer bias. Although the use of a "higher-level" test of visual recognition memory (Fagan novelty preference test) in addition to a grating detection acuity test is laudable, not all subjects were tested with the Fagan test and the methods are not clearly described. It is not clear how this study contributes much beyond the study of ACP grating acuity in handicapped children by O'Dell et al (1993) and the several studies by Hertz and colleagues. With more subjects and more thought concerning the Fagan test, the authors may be able to provide useful new information regarding visual function in multihandicapped individuals. However, the method used to test grating acuity must be clarified, and control over observer bias must be demonstrated.

1. The number of subjects is too small for useful generalization to other populations, despite being the total "deaf-blind" population in the Oslo region. In two subjects, no retest was done, therefore, test-retest reliability was based on only 10 subject.

2. No mention is made of the subjects' deafness status. Is this just a terminology usage, where "deaf-blind" does not necessarily mean deaf or blind? Patients' eye diagnoses are listed in Table 1 however, the cause of "blindness" diagnosis is unclear. In three subjects, an anomaly of one eye only is listed. Considerably more detail on the subjects' ophthalmological and neurological status would be required to adequately interpret the <u>binocular</u> acuities tested in this study. Are these subjects cortically visually impaired?

3. The TAC acuity card procedure described in this manuscript is a type of staircase and not the procedure recommended in any of the publications by Davida Teller, Velma Dobson and others. Prior to the introduction of the ACP, several groups used staircase procedures for clinical testing of forced-choice preferential looking grating acuity (including Eileen Birch, G. Mohn and Jackie van Hof;van Duin and D. Mayer and colleagues). However, the ACP was designed specifically to surmount the statistical problems of small numbers of trials in staircase or other short-cut FPL procedures (see background to development of the ACP in Teller et al (Dev Med Child Neurol, 1986; 28:779-   ), and V. Dobson's chapter in Isenberg SJ (Ed.), Eye in Infancy, 1994, St. Louis: Mosby, pp. 131-156.

4. Moreover, the staircase rule described in this paper was not one that would estimate the 70.7% threshold appropriate for a two-alternative forced-choice task. The authors' rule appears to be

appropriate for a yes-no psychophysical procedure. If the description is actually how the staircase was run, with the observer's correct and incorrect guesses on single presentations of a grating of a certain spatial frequency scored, acuity of the subjects' was likely grossly overestimated. Their staircase rule would converge on 50% correct, thus, toward higher acuities than the 70.7% threshold estimated in the 2 up-1 down rule. (For the effects of different staircase rules on threshold estimation, see Levitt H, J. Acoust Soc Am 1971; 49:476- ; for statistical issues of staircases, see Rose RM et al, Percept Psychophys 1970; 8:199- ; and McKee et al

5. The time required for testing each subject suggests that the procedure used was not an acuity card procedure. Testing should be completed in no more than 10-15 minutes.

6. Within-tester reliability was measured in this study. Most of the studies cited and compared to the results of this study were between-tester reliabilities. The authors do not make this distinction in discussing their results.

7. One observer tested subjects on both occasions. It is unclear from the methods whether the observer remained ignorant of the first test results when conducting the second test, or in what way control over the possibility of observer bias was attained.

8. In the results section, test-retest reliability of less than 0.5 oct or less is cited. However, in 3 subjects, a one step (card) difference was found, which on average is equivalent to 0.5 oct. Most other studies of handicapped and high risk infants and children have found considerably poorer test-retest reliability than that reported in this study. In one study, reliability decreased with increasing severity of disability (Hertz & Rosenberg, J Ped Opnthalmol Strab 1988; 25:139-144). This makes one suspicious of observer bias in this study.

9. Are the authors aware that Snellen (recognition) acuity may be overestimated using grating stimuli? I believe that the WHO definition of legal blindness is the one used in the US, that is, 20/200 Snellen or letter recognition acuity. The pitfalls of using grating acuities for the purpose of funding for disability services has been discussed in a recent editorial (Kushner BJ, Arch Ophthal 1994; 112: 1030-31), letter and reply (Arch Ophthal 1995; 113:970-971).

10. The authors' conclusions regarding the visual acuity status of their subjects as approaching "normal vision (sic)" (p. 9) must be considered premature, given the potential overestimation of acuity due to their staircase rule.

11. I would quibble with the author's use of the term "high-level" vision to describe the Fagan Test. How high level can it be ~~testing~~ if 5-7 month old infants are capable of this test? In fact, is not simple recognition memory an early visual processing capacity?

12. The authors tested the Fagan Test only in those subjects with "sufficiently high preferential looking acuity." (p. 11). This would seem to prejudge the results on the Fagan Test. The authors give no rationale for the use of 6.5 cy/deg as a cutoff criterion. Indeed, their value is high relative to the lower normal limits of binocular acuity in infants at an age in which the Fagan Test is conducted (e.g. 3.4 cy/deg at age 6 months in one study, Salamao et al, IOVS, 1995;36:657-670).

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12. Nov. 96

11.11.96 11:26/Prof.ZR/rt

Re: MS No. VRE2214

Dear Dr. Jacobsen,

in response to the e-mail by Sven Magnussen regarding the manuscript VRE2214 by Jacobsen et al. entiteled „Hidden visual capabilities in mentally retarted subjects diagnosed as deaf-blind" the reviewer changed his/her opinion on your paper but still has quite a number of concerns. But I now see the possibility that your manuscript might be accepted for publication in Vision Research eventually, while I cannot take a decision at this point.

I, therefore, kindly ask you to please provide a revised version that takes into account the comments of the reviewers to the extent you can follow these recommendations. When you resubmit your manuscript, please include a summary of the changes made and a brief response to all recommendations and criticisms. As soon as we receive your revised version it will be forwarded to the referees again for a second round of reviewing.

Please mail four copies of your revised version to VISION RESEARCH in Amsterdam and one copy to us.

I am looking forward to the revised version of your manuscript.

With best regards,

Eberhart [redacted]
Section Editor of VISION RESEARCH
Section 5: CLINICAL VISION SCIENCES

enclosures
referees' comments

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#1

Response to the letter from one of the coauthors of MS VRE2214 commenting on my review, forwarded to me on 8 October 1996 by editor of Vision Research.

My review was possibly too critical, and I was in error about their "staircase", for which I apologize. However, the main concerns regarding observer bias and lack of test-retest reliability data in this study remains. Possibly, clearer writing about the test methods and how observer bias was dealt with could clear up these concerns. I agree with Reviewer #2 that the introduction should be shortened considerably.

My concern in #1 was in part that many children "diagnosed" as deaf-blind are neither. In some cases it is because the diagnostician has not adequately tested the person's hearing or vision, and in others, the person may be hard-of-hearing and visually impaired. The authors know this as indicated by the title of their article. Perhaps they could keep with the "diagnosed as deaf-blind" description throughout the manuscript, especially in Method section, p. 6. The phrase, "born deaf-blind" is awkward and should be rephrased.

General comment on problem of observer bias in acuity card procedures:

In clinical practice using the Teller acuity cards, an observer will occasionally judge that a patient who in fact has bare or no light perception sees one or several gratings (usually only the few coarsest ones). Acuity also may be overestimated in normal infants and in patients with poor acuity but definite pattern vision. In normals, this occurs often by the inexperienced observer, and in patients occasionally by the experienced observer.

This is why, even in the most difficult-to-assess patients, including very low functioning patients (i.e. vegetative), confirmation of the test results by another observer is essential to establishing the validity of the test in that patient or in a study of a group of patients. There is no gold standard for a test of grating resolution acuity in pediatric patients, and one case does not make the point if there are no independent observations to confirm the finding. This manuscript may be criticized by clinicians and researchers on the point that the authors failed to provide independent validity of the test results on these patients.

However, if the observer in the study the authors' report was actually ignorant of card spatial frequency in addition to the right/left position of the grating (requiring a confederate during the procedure), as would be the case in an objective psychophysical procedure (e.g. a staircase), this could have avoided the problem of observer bias. If the test procedure was not completely objective (or if the staircase rule structure was not appropriate for the psychophysical procedure used) bias of estimates toward higher spatial frequencies is likely.

Specific questions to be addressed:

1. A staircase procedure IS NOT recommended (as stated on p. 7, line 6-7) either in the TAC manual nor by any of the acuity card developers, as far as I know. The only way I know of doing an objective staircase procedure using acuity cards requires a confederate to order the spatial frequencies, confirm the observer's judgments, and record the results.

RE: MS VRE2214, p. 2

This is not feasible in most clinical settings.

2. The exact method used in the staircase described is unclear: was a confederate involved? what were the number of trials at each spatial frequency, what were the change rules?

It sounds like it was a hybrid procedure, and if so, should be described as such. If it was a standard staircase this should be referenced. My confuson about the exact procedure they used is embedded in the following specific concerns:

a. p. 7, bottom line: which "low spatial frequency" started the test? How was this start card determined?

b. p. 8, line 3: The term "lowest spatial frequency" of 6-8 reversals is confusing. What if the reversal frequencies were not 1/2 oct (1 card) steps different but rather 1 oct (2 cards) or more oct steps different. Was the lowest SF truly selected as threshold in these cases? If the reversals always occurred between adjacent spatial frequencies (ie., 1/2 oct difference), this should be stated.

c. p. 8, line 8: "below" is a poor term here; do you mean the next highest spatial frequency?

3. The manuscript should state explicitly how much information was available to the observer regarding the previous test results. How was observer bias addressed in this study?

4. P. 7, line 9: Grating contrast IS NOT 0.5 (or 50%), but nominally 82%-84% contrast.

5. p. 8, line 11: could the authors provide the median time to test in a single session with the 11 subjects in which this was possible?

6, p. 8, line 14-15: Please state the median and range of days between tests here.

7. p. 9, line 6: This statement is in error, the test-retest differences in three patients were 0.4 oct in 1 and 0.6 oct in 2. Thus, it is more appropriate to say something like, "acuity estimates in the 10 patients who were retested differed by no more than approximately 0.5 oct." (If you calculate the card differences in the TAC set, they are on average 0.5 oct.)

8. Spelling or typographical errors on:

p. 9, line 8 ("macula") and 13 ("reminding")

p. 12, second to last line: "suggest" needs -s

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26.02.97 11:55/Prof.ZR/rt
Re: MS No. VRE2214

Dear Dr. Jacobsen,

Your paper "Hidden visual capabilities in mentally retarted subjects diagnosed as deaf-blind" (VRE2214) has been accepted for publication in VISION RESEARCH. The referee has still some suggestions for minor improvements that you might want to incorporate in the final version of the manuscript.

Please mail two hard copies and if possible a computer disk containing the final version to the central office of VISION RESEARCH in Amsterdam and one hard copy to us (instructions for electronic manuscripts enclosed).

I am looking forward to the final version of your manuscript.

With best regards,

~~Eberhart Zrenner, MD~~
Section Editor of VISION RESEARCH
Section 5: CLINICAL VISION SCIENCES

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#1

Response to revision of MS VRE 2214

The manuscript is nicely improved and I hope publication will be swift. However, I am still a little confused by the staircase scoring rule. The text says, "acuity is defined as the lowest spatial frequency of six-to-eight reversals of the staircase..." (p. 6, second from last line). Lowest means coarsest grating presented at any of the reversals, which is fact would be 3-4, as only half the reversals pertain. I suppose this rule was used because, based upon the description of the procedure, the observer presented each card only once, making a judgment of grating location on that one presentation only. This is a bit strange as the observer was not taking advantage of the opportunity to present the grating on the right and left, by 180 deg rotations of the card. This is, in fact, an essential component of the acuity card procedure and provides the observer cumulative information about the child's detection from a series of presentations of a grating.

If the acuity card procedure had been used - that is, the observer presented the card sufficiently often to judge whether the child saw the grating, thus making a Yes-No judgment on each card, the scoring rule used in this study would have been somewhat conservative. If the acuity card procedure had been used, it would have been more appropriate to obtain the geometric mean of the reversal frequencies, thus estimating the 50% Yes point on the psychometric function. In any case, the underestimation of acuity using this scoring rule is probably no more than 1/4 oct. That the scoring rule used is somewhat conservative could be stated in the discussion as evidence that it is unlikely that these children's acuities were OVER-estimated, as some readers might think.

Some minor corrections:

p. 3, line 11, "where" should be "that"

p. 4, line 7, "already" should be "previously"

p. 4, line 16, the sentence needs "acuities": "In this sense, FCPL acuities only index...."

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Hidden Visual Capabilities in Mentally Retarded Subjects Diagnosed as Deaf–Blind

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The visual acuity of twelve multi-handicapped, mentally retarded subjects, diagnosed as deaf–blind, was measured on two occasions with the Teller Acuity Cards (TAC). Eight subjects scored above the criterion for legally blind and the results of six of these indicated various degrees of poor to approaching-normal eyesight. To evaluate high-level vision four subjects were tested with the Fagan Test, assessing visual recognition memory for faces subsequent to familiarization with the preferential looking technique. The results for three subjects showed evidence for perceptual recognition. It is concluded that TAC combined with the Fagan Test may detect perceptual capacities unnoticed by clinical observation. © 1997 Elsevier Science Ltd

Visual acuity   Mental retardation   Preferential looking   Deaf–blind

INTRODUCTION

A number of reports indicate that the forced-choice preferential-looking technique (FCPL), originally developed for testing vision in infants (Teller, 1979; Mohn & Van Hof-van Duin, 1991), may be successfully applied to measure visual acuity in multi-handicapped children and adults who are not testable with conventional clinical ophthalmological procedures (Mohn & Van Hof-van Duin, 1983; Hall *et al.*, 1991; O'Dell *et al.*, 1993). The FCPL is based on the infant's tendency to fixate any visible object in an otherwise empty field, and does not require perceptual recognition, language skills or co-ordinated manual responses. When measuring visual acuity with FCPL, black-and-white luminance gratings are presented randomly to the left or right, and an observer who is blind to the grating presentation watches the subject's eye movements and guesses the location of the grating; visual acuity is the highest spatial frequency that the observer guesses the location of the grating with a probability of 0.75 correct (Teller, 1979). Mohn & Van Hof-van Duin (1983) used the preferential-looking technique to test children with neurological disorders, and were able to determine visual acuity for a majority of the subjects despite severe psychomotor retardation in some of the cases. Similar results have been reported for mentally retarded children and young adults (Lennerstrand *et al.*, 1983), pediatric patients with developmental disorders (Mayer *et al.*, 1983) and children with cortical visual impairment (Birch & Bane, 1991).

Most of these studies were laboratory based with different types of apparatus and test conditions. However, Teller and her associates have developed A standard test situation for clinical use, consisting of a set of calibrated photographic cards and a cardboard stage (Teller *et al.*, 1986). Recently O'Dell *et al.* (1993) tested 271 individuals with mental retardation with the Teller Acuity Cards (TAC). They analysed test–retest as well as intra- and inter-observer reliability, and concluded that the acuity card procedure is a useful vision screening tool for these patients. Similar conclusions were reached by Hertz & Rosenberg (1988, 1992) for mentally retarded and spastic children, and by von Tetzchner *et al.* (1996) for females with Rett syndrome.

These studies show that the varieties of the FCPL including the technically simple acuity card procedure, are valid and reliable techniques for determining visual acuity in severely retarded and disabled persons. In the present study we have used the TAC to test a selected group of subjects, namely multi-handicapped, severely mentally retarded persons who were previously diagnosed as deaf–blind.

A reliable FCPL acuity obtained in severely disabled persons does not by itself tell us anything about her/his visual perception in the conventional sense of visual recognition. It is conceivable that FCPL performance in some subjects may be governed by subcortical mechanisms of vision which are not involved in conscious perception. Studies of "blind-sight" in humans with unilateral lesions of the visual cortex (Weiskrantz, 1986; Ruddock, 1991; Stoerig, 1996) show above chance visual performance on detection and localization without conscious awareness of the stimuli, and in some of these cases the performance must be controlled by subcortical mechanisms (e.g. Magnussen & Mathiesen, 1989). In this

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TABLE 1. Subject details, visual acuity and novelty preference for faces in a sample of severely mentally retarded subjects previously diagnosed as deaf–blind

Subject details					TAC	Fagan
Subject	Sex	Age (yr)	Ophthalmological diagnosis	Refraction	Visual acuity c/deg test–retest	% Novelty preference
1	F	4	Cataract Glaucoma	+3.0, +3.5 Astigmatism	4.8–4.8	
2	F	14	Cataract RE	−2.0, RE	19.0–19.0	53.7
3	M	17	—	+5.0, +4.0	9.8–9.8	33.3
4	M	18	MaDeg	+1.0, Bin	2.4–2.4	
5	M	18	Cataract	—	0–0	
6	F	19	Cerat RE	−4.5, LE	4.8–4.8	
7	F	20	Cataract LE	−2.0, RE	9.8–13.0	57.0
8	F	23	Cataract RE Glaucoma LE	—	6.5–NR	
9	M	23	Cataract	—	1.6–2.4	
10	M	27	Cataract	+2.0, RE	6.5–NR	
11	F	28	—	Astigmatism	9.8–9.8	48.9
12	F	30	Cataract	+3.5, Bin	2.4–1.6	

RE, right eye; LE, left eye, MaDeg, macula degeneration; Cerat, ceratoconus; Bin, binocular; NR, not retested.

sense FCPL acuities only index the resolution of the sensory system, and do not tell us about high-level vision (Van Hof-van Duin, 1989; Mohn & Van Hof-van Duin, 1991). To conclude that the person has functional vision some additional indication of pattern or object discrimination is required. In the present paper a subsample of the subjects was tested with the Fagan Test, which is a preferential looking test of visual recognition memory for faces (Fagan & Detterman, 1992).

PREFERENTIAL LOOKING ACUITY

Method

Subjects. The National Health Department keeps a continuous record of deaf–blind persons living in Norway; in 1994 a total of 86 persons diagnosed as born deaf–blind were registered of a population of about four million people. In the present study, 12 mentally retarded children and young adults from that register were tested. The sample comprised 14% of this population in Norway, and all the persons diagnosed as born deaf–blind living in the Oslo region where the study was conducted. Six of the subjects were females and six were males, varying from 4 to 30 years of age (Table 1). Using the criteria of DSM-IV (American Psychiatric Association, 1994) seven subjects were diagnosed as severely mentally retarded (subjects 1, 2, 4, 7, 8, 10, 11) and five as profoundly retarded (3, 5, 6, 9, 12). All subjects were multi-handicapped in the sense that they had combined sensory impairments; in addition, the subjects diagnosed as profoundly retarded exhibited severe psychomotor retardation. The subjects were unable to communicate in any formal language, hand-language or otherwise, except for subjects 2, 7, 10 and 11, who were able to signal very basic needs.

In Norway, assessment and diagnoses of the visual capacities of the mentally retarded deaf–blind persons are based on a combination of ophthalmological examination, careful clinical observation and informal testing of attention and responsiveness to visual stimuli, carried out by trained personnel. Ophthalmological data were available from the medical records of all the subjects in the present sample, and are summarized in Table 1. Nine subjects had refractive errors and these subjects were fitted with lenses prior to testing, except for one subject (No. 4) who refused to wear spectacles.

Procedure. Visual acuity was measured with the Teller Acuity Cards with a modified test administration which included forced choice judgements, blind testing and a variant of the psychophysical staircase procedure. The TAC consists of 15 neutral grey cards containing a 12.5×12.5 cm square-wave grating at a distance of 7 cm from a tiny peephole in the centre; grating contrast is about 80% and background luminance matches the space average luminance of the grating. The spatial frequency varies between 0.32 and 38.0 c/deg for a viewing distance of 55 cm. The acuity cards are presented in a rectangular window in a larger cardboard screen shielding the subject's visual field.

Eight of the subjects were tested in either home or institutional settings, four subjects were tested at a laboratory at the university. The subjects were either sitting in a supporting chair or, when not possible, were tested lying down resting on a pillow. In these (three) cases the cardboard screen had to be abandoned, but distance markers and lighting conditions were improvised so that in all cases test distance and card luminance conformed to test requirements. Testing was binocular.

When the subject was considered attentive by the observer, a card with a grating randomly to the right or left was presented. The observer, who did not know the location of the grating, observed the subject's eye movements through the peephole in the centre of the card and guessed the location of the grating. Presentation always started with the lowest spatial frequency card and proceeded in half-octave steps towards higher spatial frequencies: when the observer's guess was correct a higher spatial frequency was chosen on the next trial, when the observer's guess was wrong, a lower spatial frequency was chosen, always in half-octave steps. In the present study acuity is defined as the lowest spatial frequency of six-to-eight consecutive reversals of the staircase involving adjacent spatial frequencies. To achieve blind testing and avoid observer bias, the test cards were arranged on a table from low to high spatial frequencies with the grating patterns facing down; the observer rotated the card several times, both before presentation and upon replacing the card on the table—thus he had no clue to the location of the grating on any trial.

In several cases it was not possible to catch the subject's attention over a sufficiently long time to complete testing in a single run. In this case the test was run a second time after a suitable break, starting once again with the lowest spatial frequency card and then proceeding directly to the next highest spatial frequency producing an correct guess by the observer on the previous run. With a flexible procedure and suitable improvisations visual acuity could be determined for 11 subjects in a single session lasting from 20 to 80 min; one subject (No. 5) showed no response to any test card in either test or retest sessions.

Except for two subjects who moved to another part of the country shortly after the first test session, subjects were tested on two occasions with a minimum interval of 20 days between test and retest (median test–retest interval 27.5 days, range 20–30 days). All tests were performed by an experienced observer (author KJ). Individual scores on the first session were not consulted prior to retesting.

Results and discussion

Visual acuity estimates were obtained in 11 subjects, one subject (No. 5) with binocular cataract did not respond to any test card during either test or retest sessions. The results are shown in Table 1, with subjects assigned numbers according to age, for test and retest sessions independently. The test–retest reliability is good, confirming previous studies (Hertz & Rosenberg, 1988, 1992; O'Dell *et al*., 1993), with acuity estimates in the ten patients who were retested differing by no more than approximately 0.5 octaves.

For three subjects (Nos 4, 9 and 12) blindness was confirmed in the sense that the visual acuities measured by TAC corresponded to the legal definition of blindness proposed by WHO (spatial frequency of 2.4 c/deg, Snellen acuity of 0.1). The remaining eight subjects all exhibited visual acuities above this criterion, and in two of the subjects (Nos 2 and 7) TAC acuities approached normal vision; the visual acuity scores were generally higher in subjects diagnosed as severely retarded compared with the profoundly retarded subjects. To get an idea of the visual performance of the subjects, consider that a visual acuity of 6–8 c/deg corresponds to the acuities measured in the 6-month-old baby (Mohn & Van Hof-van Duin, 1991), the rod monochromat (Greenlee *et al*., 1988) or the adult domestic cat (Crawford *et al*., 1990). Thus, provided vision is functional, these patients are by no means blind.

The results support a number of previous studies (Hertz & Rosenberg, 1988, 1992; O'Dell *et al*., 1993; von Tetzchner *et al*., 1996) that the technically simple TAC procedure is a valid and reliable technique for determining visual acuity in severely mentally retarded subjects. However, as noted above, FCPL grating acuity tests do not index high-level vision in infants or disabled persons (Van Hof-van Duin, 1989; Mohn & Van Hof-van Duin, 1991). They do not tell us about perception proper, how the subject will perform on normal visual tasks involving recognition and identification. Conventional recognition tests (optotype tests) on the other hand, which do involve these higher-level perceptual–cognitive functions, confound problems of visual resolution, high-level vision and cognitive–communicative skills when applied to such persons. Therefore, when deficits in perception are suspected, a preferential looking test should be supplemented with a perceptual test measuring visual recognition and vice versa. However, most tests of recognition require that subjects understand and can follow verbal instructions and make an appropriate response. They are, therefore, not applicable to the current subjects.

To evaluate perceptual recognition we decided to test those subjects who had a sufficiently high acuity score with the Fagan Test (Fagan & Detterman, 1992), recently applied to evaluate the presence of high-level vision in Rett syndrome (von Tetzchner *et al*., 1996). The Fagan Test is a test of visual recognition memory for faces in a variant of the preferential looking procedure: on a given trial two identical pictures of a face are first presented for inspection, this stimulus is then paired with another face, and the subject's preference is measured by looking time. If the subject systematically favours either the familiar or the novel face, perceptual discrimination must be present.

FACE DISCRIMINATION

Method

Face discrimination was tested by the computer edition of the child and adult version of the Fagan Test for age 2 years and above (Fagan & Vasen, 1997). Six of the subjects in the first study were judged to have sufficiently high preferential looking acuity to be tested with the facial patterns (6.5 c/deg or better, subjects Nos 2, 3, 7, 8, 10, 11) but two of these (Nos 8 and 10) moved to another part of the country before the Fagan Test was available, leaving four subjects with acuities of 9.8 c/deg or better, three severely retarded and one profoundly retarded patient. This criterion was chosen to avoid confounding negative results on perception recognition with poor visual acuity; in addition, the Fagan Test is computerized, which poses certain practical limitations on testing.

The Fagan Test consists of 20 novelty tasks, all involving comparison of achromatic pictures of human faces presented on a computer screen. There are two blocks of 10 trials, one testing immediate memory and one testing delayed memory. In the immediate memory condition each trial consists of a 5-sec presentation of a single face, immediately followed by a 4-sec test presentation of the "familiar" face paired with a novel face which switch left–right position after 2 sec. In the delayed memory condition, the novel face is presented together with a face shown on one of the previous trials in the first block. At a viewing distance of 55 cm the computer screen subtended 30 deg visual angle, the face stimuli subtended 13 deg visual angle, and they were separated by a 4 deg blank space.

Visual recognition is measured by the differences in looking time between the novel and the familiar face. The observer, hidden behind a cardboard screen observes the corneal reflections of the targets through a tiny hole, and records left or right looking by pressing one of two mouse buttons. Testing was performed by a well-trained observer (author LS). Average inter-observer reliability of trained observers in recording differential fixation is 0.94 (Fagan & Detterman, 1992).

Results and discussion

The right column in Table 1 shows preference scores on the Fagan Test in terms of accumulated looking time for new faces in per cent of total looking time. The standardization of this test defines 47/53 per cent criterion for displayed preference (Fagan *et al.*, 1986). Three of the four subjects scored beyond this limit, with two subjects (No. 3, profoundly retarded, and No. 7, severely retarded) exhibiting a strong preference for either the familiar or novel face (33.3 and 57.0% looking time) and one subject (No. 2) exhibiting moderate preference for the novel face (53.7%). One subject (No. 11) showed no preference in either direction according to the test criteria (48.9%).

The results indicate that three of the subjects possess the perceptual–cognitive skills necessary to discriminate and remember human faces, whereas for one subject such skills have not been demonstrated. Note, however, that the logic of the test only permits affirmative conclusions. Strictly speaking, chance performance on the Fagan Test does not imply that perceptual discrimination is not present, only that it does not turn up in a systematic across-trials perceptual preference pattern. However, the absence of face preference is an indication that the subjects do not possess functional vision despite a moderate visual acuity score on the TAC.

GENERAL DISCUSSION

The results of the present study show that with suitable testing procedures it is possible to obtain evidence for perceptual capacities that were otherwise bypassed by clinical observation. With the TAC procedure visual acuity could be determined in 11 of 12 severely or profoundly mentally retarded children and young adults that had been diagnosed as deaf–blind by the medical examination routines of the Norwegian health care system for multi-handicapped people, and eight of these proved to possess various degrees of poor to approaching-normal visual acuity. While preferential looking acuity does not by itself prove the presence of perceptual processing, tests of visual recognition carried out on four of the subjects gave evidence for perceptual recognition in three subjects, and we might suspect similar capacities in some of the subjects who were not tested. Since the Fagan Test consists of two-stimuli cards, perceptual preference must be based on discrimination of facial identity or facial expression. This type of perceptual performance is beyond blind-sight mediated by subcortical structures (Weiskrantz, 1986; Ruddock, 1991; Stoerig, 1996), and indicates the presence of genuine conscious perception. While the TAC results sugggest overall better acuities among severely retarded compared with profoundly retarded patients, the single patient in the latter category tested with the Fagan Test showed clear evidence of perceptual recognition. However, our sample is too limited to allow any conclusions about the relationship between the level of mental retardation and visual performance.

Why were perceptual capacities not previously detected in these subjects? The main factor of course is that these patients are very difficult to test by any type of test, and since multi-handicapped subjects frequently are passive, lack initiative and do not respond to visual stimuli, their behaviour might be misinterpreted as a sign of functional blindness. Passivity is typical for blind children (Burlingham, 1975; Fraiberg, 1977), but may also be a consequence of, for example, disturbed motor functions (von Tetzchner *et al.*, 1996), cognitive dysfunctions (Mundy & Sigman, 1989) or even emotional disturbances (Beck *et al.*, 1992; Wells & Mattews, 1994). More informal observation procedures and clinical judgement may fail to identify the cause of passivity. Forced-choice preferential looking techniques which do not rely heavily on higher-level cognitive processes or motor responses would seem to be an ideal tool evaluating the sensory capabilities of these patients. Combining the TAC with some measure of visual discrimination, such as the Fagan test, we can further decide if the visual communication channel is open or closed. In order to develop efficient habilitation programmes for multi-handicapped persons it is necessary to be able to differentiate between sensory deficits and other factors limiting interaction and behavioural performance. TAC and the Fagan test should both be useful tools in this process.

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