A STUDY OF DYSLEXIA

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The syndrome of "pure alexia", i.e., alexia without aphasia or agraphia, was first recorded in this country by Broadbent (1872) and a number of cases have since been described in the literature. Adequate historical accounts have been given by de Massary (1932), Weisenberg and McBride (1935), and Russell Brain (1955). In recent years, fresh interest in the syndrome has been aroused by the excellent study of Holmes (1950). Its possible relevance to the problem of specific reading disability in children (Ettlinger and Jackson, 1955) provides additional grounds for further inquiry.

Although "pure alexia" is widely accepted as a clinical entity, it has often been pointed out that the term is a misnomer. In the first place, the defect of reading is commonly, if not invariably, associated with other visual deficits. Apart from right hemianopia (which, with one doubtful exception, has been reported in every published case known to us), an apparently specific defect of colour recognition is almost invariably linked with alexia (Poezl, 1928; Stengel, 1948). Some degree of visual-spatial agnosia or disorientation has also been claimed by several authors (Wolpert, 1930; Martin, 1954). In the second place, minor grades of dysphasia, dysgraphia, and dyscalculia can be traced in the majority of published cases (Weisenberg and McBride, 1935) and may in the opinion of many invariably be elicited on careful examination (Critchley, 1953). None the less, the predominance in certain cases of reading disability without appreciable involvement of other functions, visual or linguistic, would appear an established fact (Brain, 1955). To this extent at least, alexia may be said to present as a circumscribed psychological deficit.

The cerebral localization of alexia is a matter of some interest. Whereas the locus of the lesion provoking this syndrome was for long held to be the left angular gyrus (Dejerine, 1914), more recent work has consistently implicated the basal parts of the occipital lobe, in particular the lingual and fusiform gyri. Some damage to callosal fibres has also been adduced in the majority of cases verified by necropsy (Kleist, 1934; Gloning, Gloning, Seidelberger, and Tschabitscher, 1955). It is also noteworthy that alexia without other signs of visual agnosia has been reported regularly to follow left occipital lobectomy (Hécaen, Ajuriaguerra, and David, 1952). In its more circumscribed forms at least, alexia would clearly appear to be a syndrome of the dominant occipital lobe.

It has been traditional to regard alexia as a highly selective variety of visual agnosia, i.e., a loss or defect of recognition limited to linguistic symbols (though numerals are often excepted). Russell Brain (1955), for example, considers the basic defect to lie in a failure on the part of the perceived material to arouse the visual word-schemes ("memory traces") which may be assumed to mediate recognition of words and evocation of their meanings. Although this view commands wide acceptance it has not gone unchallenged. As long ago as 1930, Beringer and Stein attempted an explanation of alexia in terms not of a symbolic defect but of an abnormal functional lability (pathologische Funktionswandel) of the visual system. This lability was held to produce what is in effect a concentric constriction of vision leading to grossly impaired perception of fine detail.* In more recent years a similar explanation has been repeatedly advocated by Bay (1950, 1953) with reference to agnosia in general. It is pertinent to note that Martin (1954) has also laid stress on sensory deficits, in particular visual disorientation, as causal factors in alexia. One may comment that although the role of visuo-sensory impairment in alexia may well be appreciable, the existence of a specific and localized central defect of recognition can certainly not be ruled out. Indeed cases such as that of Holmes (1950) render explanation in terms other than symbolic loss extremely difficult to contemplate.

In view of the important part played by eye movements in reading, it has been suggested that some aspects of dyslexia may be traced to derangement in the oculo-motor sphere. Thus, a defect of fixation

*Unfortunately, the relevant psychophysical data upon which this explanation was based were never published and are not now available (Hassler, personal communication, 1953).
was adduced by Holmes (1950) as a contributory factor in his own case. More recently, the relation of oculo-motor to alexic disabilities has been fully discussed by Gloning and others (1955). It remains uncertain, however, whether the abnormality of eye-movement control often seen in cases of reading disability is primary or secondary to the dyslexia. One of the main purposes of the present study is to throw further light on this problem.

We propose in this paper to communicate a case of "word-blindness" (verbal alexia) due to a left occipito-parietal meningioma in an intelligent right-handed man. The defect of reading was the main presenting symptom and a relatively persistent postoperative disability. Although some defect in fields of performance other than reading was established on careful examination, it may be doubted whether this alexia was any less "pure" than in many earlier cases to which "purity" has been ascribed. In addition to routine psychological examination, an experimental analysis of certain aspects of visual function was undertaken and photographic records of eye movements will be presented. It is hoped that the findings may throw light on the genesis of alexia in its more circumscribed forms.

Case Report

N.H. No. 63273.—T.S., an electrician aged 41, was admitted under the care of Dr. R. W. Gilliatt with complaints of blackouts following a head injury in 1945, and of inability to read of recent onset.

His blackouts had begun nine months after the head injury and had continued at intervals of about one per month for two to three years. He had had none in the past six years. At the beginning of each attack, he would see a black shadow moving to and fro on his right-hand side; this movement became faster and faster and he would then lose consciousness for 15 to 20 minutes. He was not incontinent in these attacks and had never bitten his tongue or otherwise injured himself.

The patient first noticed difficulty in reading about nine months before admission. This became progressively worse and the patient was now virtually unable to read. He stated that although he could see the words clearly, they did not mean anything to him unless he spelled out the letters one by one. Recently, he had had difficulty even in recognizing the individual letters themselves. Words were seen every day or in a familiar context, e.g., "electricity", were easier to read than others. The patient reported no difficulty in recognizing numerals and no defect of calculation. He stated that he could write fluently but had noticed some trouble with spelling during the past eight months, affecting both long and short words alike. He had had no difficulty in speaking or in understanding things said to him and denied ever being at a loss for names. But he had become somewhat absent-minded in recent months, e.g., forgetting where he had left his tools.

On admission, the patient was alert, cooperative, and fully orientated in all spheres. He was adequately informed about current events and expressed himself fully with a good vocabulary. Apart from some reasonable apprehension regarding his symptoms, emotional reaction was normal and appropriate. He was fully right-handed.

On neurological examination, there was a complete right homonymous hemianopia to confrontation, apparently splitting the macula. V.A.R. JI 6/9; V.A.L. JI 6/9 (individual letters). The right fundus was normal; the left showed papilloedema, with one small haemorrhage on the disc. The pupils were equal, reacting briskly to light and on convergence. External ocular movements were full with no squint or diplopia. No nystagmus was noted or other abnormality in the cranial nerves. In the limbs, no wasting, fasciculations or abnormal movements were seen; tone, power, and coordination were normal throughout but tendon reflexes were brisker right than left and both plantar responses were flexor. Pain, touch, and temperature were appreciated normally in all areas; fine movements of fingers and toes were accurately perceived and two-point discrimination on finger-tips and toes was unimpaired. Vibration sense was normal. He had no finger agnosia, but slight hesitancy regarding the right and left sides of his own body. No apraxia was noted.

Language Functions.—Speech was fluent and on no occasion was the patient hesitant or at a loss for a word. Grammar and syntax were appropriate and there was no paraphasia. Comprehension of spoken speech showed no obvious defects and even complicated oral commands were executed faultlessly. In naming objects, there was occasional hesitancy, perhaps suggesting a minimal nominal defect.

Reading was grossly affected. Individual printed letters could as a rule be recognized though occasional mistakes were made. In some cases, a letter was recognized only after the patient had traced its outline with the tip of his finger or the point of a pencil ("Wilbrand's sign"). Short words were as a rule read correctly; though slowly, and were often spelled out aloud or silently before they could be pronounced. Longer words gave much greater difficulty and these the patient could not always read correctly letter by letter. There was some evidence that increasing the size of the print rendered its comprehension easier. Numerals were read correctly. Some hunting movements of the head were observed in reading numerals though none while reading words and phrases. The patient was able to read the time. Connected prose, e.g., newspaper items, could scarcely be read at all, the patient stating that he rarely read more than one word at a time and usually had to spell words out before becoming aware of their meaning. On the other hand, he would occasionally recognize a word immediately, even a long one. Individual words were sometimes misread, and it is noteworthy that almost all the errors made were with reference to word-endings (this is analysed more fully below).

The following text from a newspaper is a sample of the patient's oral reading:—

A girl who married her employer, a bakery manager,
six months after breaking her engagement to a lorry driver, had led the jilted man “up the Registrar’s garden path”.

It was read as follows:

A girl (quickly)—(pause)—who married her employer (long pause—I can’t make that word out—the manager (pause) six weeks—six weeks after—the manager six weeks after—six weeks after breakfast-she—her engagement to a lorry driver led her a jilt—a man—is it a man?—“no”. I think it’s at the Registration—at the Registration Garden Path (that’s what I think it is—I just can’t make out that one, I just can’t make sense of it you see).

(Total time taken 12 minutes.)

It was noticed that words presented in familiar settings could often be read quickly and accurately, e.g., the name of a daily newspaper or a popular brand of cigarettes. Occasionally, however, mistakes were made, as when the patient recognized immediately a familiar advertisement of a brand of petrol but misread “benzole” as “pool”. Punctuation signs and idiolects (road signs) were correctly understood.

Asked to describe his difficulty in reading, the patient said: “I can see the words but I don’t get what I’m looking at—I can see the various letters and that but while I’m looking at them I don’t know what they are”.

When asked to fixate the first letter of a 13-letter printed word, the patient stated that he was unable to see the end of it. This he could do only when his fixation was directed to the ninth or tenth letter. His “span” for simultaneous perception of printed letters without shift of fixation appeared to be roughly seven to the left and three to the right of the fixation point.

In writing, the patient showed moderate disability. Spontaneous writing was slow, with rather poor alignment of words on the paper (Fig. 1). There was occasional difficulty in forming letters and in a few cases wrong letters were used, e.g., “py” for “by”, “ti” for “to”. In general, however, spelling was adequate, both written and oral. The patient was able to write the letters of the alphabet in capitals with only an occasional error, but had much greater difficulty in doing so in script. Writing to dictation was halting, with occasional mistakes, and copying of printed material appeared even more impaired than spontaneous writing or dictation. In particular, copying from print into cursive script gave great difficulty and was accomplished extremely slowly. On the other hand, geometrical designs could be copied or drawn from memory without gross defect.

Calculation was impaired. Although the fundamental rules of arithmetic were understood, mental arithmetic was slow and often inaccurate. The patient could not perform the 100-7 test. Arithmetic signs were as a rule read correctly, but the ± and = signs were sometimes confused.

Visual Perception.—As has been said, there was a complete right homonymous hemianopia to confrontation. Perimetry with a 2/330 test object suggested that
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Fig. 2.—Campimetric visual fields.

there was some macular sparing in the left eye, but this finding was thought to be due to defective fixation (Dr. R. W. Gilliatt). Campimetry undertaken several weeks post-operatively left no doubt whatsoever that the maculae were in fact split in both eyes (Dr. G. S. N. Russell). The campimetric fields are given in Fig. 2.

The patient was tested with a series of procedures designed to assess the functional efficiency of the perimetrically intact areas of the visual fields. These procedures have been described elsewhere (Eitlinger, 1956). All tests were applied to the left eye only. On a test of local brightness-adaptation (adapted from Bay, 1950) it was found that the patient’s “fading-times” were within the normal range at 5° but above average at 15° and 35° suggesting a slightly diminished adaptation rate. (According to Bay (1953), increase in adaptation rate is to be anticipated.) On a test of peripheral visual acuity, the findings were within normal limits. On a brightness-discrimination test, the findings indicated some loss of brightness discrimination in foveal vision, but normal results when the stimuli were presented at visual angles of 2-5°, 5°, 15°, and 35° in the left half-field. The results of these tests do not suggest significant loss of visual efficiency in the perimetrically intact regions of the left eye. It can be assumed that the findings in the right eye would be essentially similar.

Colour perception was anomalous. It is possible, however, that there was a pre-existing colour weakness, probably a deuteranopia. On examination with the Ishihara test, the patient was found to be able to read only three of the 28 items, despite the fact that his recognition of numerals was not affected. He did not give the characteristic pattern of any recognized type of congenital colour anomaly.

The patient reported no difficulty in recognizing objects, faces, or buildings and no evidence of object agnosia was forthcoming on special tests. On the other hand, pictures were apt to cause difficulty, irrespective of their size. Although individual objects were usually perceived correctly, the patient was often unable to appreciate the nature of intention of the scene depicted. He failed to grasp some rather elementary political cartoons in the daily press and could not follow strip cartoons. He missed one or two items on the very simple “mutilated pictures” sub-test at the 6-year level of the Stanford-Binet scale.

On tachistoscopic tests, the patient’s perception of forms and letters exposed for short intervals in the intact left central field was much retarded though not grossly impaired.

The patient presented no visual disorientation in any plane of space. He was able to join scattered dots by straight lines without hesitation or error. Drawings of geometrical forms or common objects, though crude, showed no gross disorganization. On the Kohs blocks, performance, though poor, was not grossly defective. On the other hand, the patient found great difficulty with the “block-counting” sub-test at the 10-year level of the Stanford-Binet scale, only half of his responses to the 14 items being correct.

Topographical orientation and memory were not obviously affected.

On formal psychometric testing, his score on the Wechsler-Bellevue vocabulary indicated a premorbid I.Q. in the region of 105. There was significant impairment on block design and arithmetic, though not on similarities. Simple memory tests (verbal and visual) were poorly performed. On the other hand, Weigl’s sorting test was executed faultlessly.

On E.E.G. examination, the small alpha rhythm at 8-9 c./sec. was diminished on the left side. Throughout this side, though maximal anteriorly, there was nearly continuous 6-8 c./sec. rhythm and, intermittently, waves at 2-3 c./sec. with sharp phases, showing a focus in the fronto-temporal region. The record was not altered on overbreathing and flicker stimulation was not tolerated. In the light of these findings, it was suggested that a large area of the left hemisphere was involved in the disturbance, with the probability of a lesion deep in the hemisphere (Dr. W. A. Cobb).

Arteriography and ventriculography suggested a large left occipito-parietal space-occupying lesion, almost certainly a menigioma. At operation (Mr. Wylie McKissock) a large left occipito-parietal neoplasm, measuring at least 10 cm. from before, backwards and extending out 8 cm. from the midline, was removed.
confirmed the movements, the Pathological successfully. Pathological examination of the specimen confirmed the diagnosis of menigioma.

Post-operative Progress.—The patient's post-operative progress was very satisfactory. One week after operation, speech was fluent: the patient could name a wide variety of objects and respond correctly to spoken commands. He had no finger-agnosia, apraxia, or disorientation for left and right. The patient could write his name and address, and print the alphabet, but was unable to write a letter. Copying of print was even more impaired than before operation. Thus, asked to copy the phrase "Give me the tools to do the job" (which the patient wrote from dictation with but a single error), all he could produce was "GIVG". His reading disability appeared to be unchanged.

Two weeks after operation, the patient was reading much better and missing fewer words. He still spelled words out to himself, "to make sure I have the right answer" but this now seemed more a matter of confirmation than of recognition itself. Errors made in reading lists of single words at this time were observed consistently to involve the end of the word and beginning. Examples are "because" for "beware", "labour" for "labourers", "projectionist" for "projecting", "water" for "waste", "terrier" for "terror", "together" for "tongue", and "oblong" for "obtained". The patient seemed aware of this tendency to misread the ends of words, and stated that his reading disability now appeared to be due more to this factor than to failure to understand meaning. He claimed that he could never see more than three or four letters of a word at the same time.

At this time, the patient reported some momentary formed hallucinations in his right visual field (e.g., an electric motor or an arm-chair), but without loss or defect of consciousness.

During the post-operative period in hospital constant help and practice with reading were given. The patient
was specifically instructed to compensate for his hemianopia by lateral head and eye movements along the line of vision and given systematic practice in doing so.

Four weeks after operation, it was found that words of any length could now be read more quickly, though still well below normal reading speed. Occasional errors were still made with word endings. Connected passages could now be read aloud quite well, though with some omission of small words or of the last letter or syllable of a longer word, e.g., “distinguish” for “distinguishable”. It was observed that the patient invariably made short, fast head movements while reading. He stated that he now never completely failed to recognize a word.

At this stage, writing had much improved but the difficulty in picture interpretation was still present. The patient failed to grasp simple pictorial jokes or cartoons and occasionally failed to recognize outline drawings of animal or human figures. Colour perception also remained defective. Tested with the Holmgren wools, the patient often failed to select a hue named by the examiner, stating that he could not remember its appearance. When asked to guess, his guesses were seldom appropriate. Naming of colours was inconsistent, with some tendency to react to brightness rather than hue (e.g., light blue was often called white or grey, pink was called pale grey, dark green was called black). The patient was quite unable to sort colours according to categories of hue.

Seven weeks after operation, it was noted that the patient’s hemianopia was unchanged. Reading and writing had improved further, but both remained slow. Re-examination 12 weeks after operation indicated a slight regression in the patient’s reading capacity. At this time he had been discharged from hospital and was no longer having daily reading practice.

Eye Movement Studies—Eye movement records taken from the patient while reading short standard texts were obtained three, seven, and twelve weeks post-operatively by means of the “opthalmograph” (American Optical Co.). All records were found to show gross impairment of the normal pattern of saccadic scanning movements and fixations. In the records taken three weeks after operation, there was scarcely any evidence of fixations at all, the motions consisting of a slow progression of the eyes along each line of print with small irregular backward and forward twitches, many of which lacked the step-wise character of the normal saccadic movement (Fig. 3a).

The records taken seven weeks after operation were found to be more normal, with some appearance of a staircase pattern (Fig. 3b), though scanning of each line was more protracted and the number of fixations considerably higher than in comparable records obtained from a patient with a right hemianopia (with central sparing, due to a left parietal gunshot wound in whom there was but mild dyslexia (Fig. 3c), and from a healthy adult subject (Fig. 3d).

The records obtained after 12 weeks indicated some slight deterioration of eye movement responses as compared with those obtained seven weeks after operation. As these records were technically imperfect they are not reproduced here.

Discussion

This is a case of "word-blindness" (without appreciable "letter-blindness") associated with a large left occipito-parietal meningioma in a right-handed man. Writing was but little affected, except during the immediate post-operative period, and there was no apparent defect of oral comprehension or spoken speech. The patient presented a complete right hemianopia, splitting the macula, and some weakness of colour vision probably due in part to pre-existing deuteranopia. Apart from slight difficulty in interpreting pictures, there was no real object-agnosia or disturbance of spatial perception. Visual orientation, in particular, appeared fully intact. Nor was there appreciable change in local adaptation or discriminative sensitivity to form and brightness in the intact right half-fields of vision. Photographic eye movement records obtained three weeks post-operatively showed gross abnormality. There was no apraxia of constructional defect, apart from a curious and apparently selective difficulty in copying print, as in the case reported by Holmes (1950). Except for some slight memory impairment, the overall intellectual level was well preserved throughout.

The patient’s reading gradually improved after operation with an intensive course of re-education. It is noteworthy that this improvement appeared to be correlated with increasing awareness of the hemianopia and the appearance of compensatory lateral head and eye movements in reading. In keeping with this improvement, photographic eye-movement records obtained seven weeks after operation were found to present a more normal pattern. One may therefore inquire what (if any) relation there may be between defects in oculomotor control and this unusually circumscribed alexia.

It has long been known that disorder in oculomotor coordination and control may present from cerebral lesions (Holmes, 1938). In the present case, however, there was little to suggest a primary, executive derangement of coordinated eye movements. Fixation was adequate and pursuit movements of the eyes did not appear grossly ataxic. It is therefore probable that the abnormal character of the eye-movement records in this case is to be interpreted as secondary to a disorder of perception. Thus it is easy to suppose that the normal sequence of eye movements in reading was slowed and at times disrupted by the patient’s failure to grasp the sense of what he read. This is of course the most obvious explanation and we certainly should not wish to discount it. At the same time, our analysis
of the patient’s reading disability has suggested to us that factors other than agnosia in the traditional sense may well have contributed to its origin. As such factors do not appear to have been previously considered in connexion with alexia we may devote some discussion to them.

This patient presented a complete right hemianopia without macular sparing. In the earlier stages, moreover, he appeared essentially unaware of this field defect and there was no evidence that he had compensated for it either by the elaboration of a “pseudo-fovea” (Fuchs, 1922) or by the use of small-range lateral eye movements (Critchley, 1953). Indeed, the degree of neglect of the hemianopia was so pronounced as to suggest unilateral imperception or denial of disability (Critchley, 1953). At all events, it appears reasonable to assume that the patient’s capacity to explore the right half of the visual world by means of appropriate eye movements was seriously defective. In reading, it would appear that he at first made no allowance for his field defect and that stimuli whose images fell to the right of the fixation point in consequence failed to elicit a normal, quasi-reflex, saccadic eye movement leading to a fresh fixation. That is to say, the reading disability arose, in part at least, from a failure in oculomotor adjustment secondary to a complete right homonymous hemianopia.

The above explanation may be held to account for several unusual features of the patient’s reading disability. Thus it will be borne in mind that he was almost always able to read single letters and short words, which did not necessitate a shift of fixation. Further, his errors in reading longer words or phrases almost without exception bore on the word endings, suggesting a failure to scan the entire word or line. It is also noteworthy that the improvement in reading in the post-operative period appeared to be correlated with gain of insight into the hemianopia and the compensatory development of voluntary lateral head and eye movements. Taken together, these findings suggest that an important element in the patient’s dyslexia was a failure in systematic lateral eye movement from left to right with consequent defect in continuous reading.

It is, of course, well established that not every case of right hemianopia presents difficulty in reading comparable with that observed in the present case. Yet it is noteworthy that left occipital lobectomy, which is known to produce right hemianopia without central sparing, is regularly followed by circumscribed alexia (Hécaen and others, 1952). It is also relevant to note that “pure alexia” is seldom encountered in patients with sparing of the macula and good awareness of the field defect. In such cases, moreover, photographic records of eye movements in reading may present a far less abnormal appearance (cf. Fig. 3c). Although the relations between field defects, oculomotor control, and alexia obviously require further elucidation, it is at least possible that a combination of defects of the kind we have described may be responsible for the apparent “purity” of the alexia in many earlier cases reported in the literature.

At the same time, we certainly should not wish to claim that an explanation along the above lines will account for all aspects of the reading disability in this case. In view of the presence of correlated, if less severe, defects in copying, writing, picture interpretation, and calculation, the possibility of a central weakness in the recognition of visual symbols can certainly not be excluded. Nor would we wish to claim that cases of alexia in which the defect extends to the recognition of individual letters, as in that of Holmes (1950), can be explained along these lines. In such cases, it appears necessary to postulate a selective agnosia, possibly consequent upon a defect in visual orientation as suggested by Martin (1954). We wish to do no more than call attention to the significance for reading of defective lateral eye movement consequent upon a complete right-sided field defect with unrewardedness of hemianopia. The extent to which this combination of handicaps may constrain alexia in general must await the outcome of further study.

Summary

The syndrome of “pure alexia” is briefly discussed.

A case is reported in which gross defect in reading (verbal alexia) was associated with a left occipito-parietal meningioma in a right-handed man. There was a complete right hemianopia but no loss of differential form or brightness sensitivity in the intact left half-fields. The dyslexia was associated with mild dysgraphia and dyscalculia but with no defect of speech, comprehension, or spatial judgment. Colour perception was anomalous.

“Ophthalmograph” eye movement records obtained post-operatively are presented. These reveal gross derangement of the normal pattern of scanning movements and fixations in reading. The relation of this finding to the right-sided visual field defect is considered.

The relative parts played by visual field defect, oculomotor derangement, and loss of recognition of visual symbols in the genesis of alexia are discussed. It is concluded that the outspoken character of the alexia in this case was due to an uncompensated right hemianopia with resultant derangement of the oculomotor scanning mechanism.
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