A CAPILLAROSCOPIC INVESTIGATION
OF MONGOLISM

by
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A THESIS SUBMITTED IN PARTIAL FULFILMENT OF
THE REQUIREMENTS FOR THE DEGREE OF
MASTER OF Science
in the Department
of
Neurological Research

We accept this thesis as conforming to the
standard required from candidates for the
degree of MASTER OF Science.

Members of the Department of
Neurological Research

THE UNIVERSITY OF BRITISH COLUMBIA.

October, 1957.
The purpose of the study was to determine the morphological capillary pattern of the mongoloids and compare this with another constitutionally retarded clinical group - the phenylketonurics - and the degree of deviation from the "normal" pattern.

Photomicrographs were taken with capillary microscope and the capillaries were analysed into the ten basic morphological categories as set out by Gibson, Bosley and Griffiths (1956).

It was concluded that the mongoloids and the phenylketonurics differed significantly in the amount of primitive immature configuration they displayed. The hypothesis of Powdermaker (1929) that structure is related to function, was found tenable through a capillaroscopic examination of scholastically retarded school children.
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Date September 12, 1957.
ACKNOWLEDGEMENTS

The writer wishes to thank Dr. W.C. Gibson for his skillful guidance in conducting the research project, and also Dr. J. Wada for his constructive criticisms of the thesis. Special thanks are extended to the nursing staff at Woodlands School who offered a great deal of assistance in taking the photographs of the patients. Also to Dr. Kerwood who recognized the importance of carrying out basic research and made the services of the school available.
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CHAPTER I

INTRODUCTION

Following the monumental work of Itard, the French physician, who developed a system for the education of defective children, there sprang a new interest centred on mental deficiency. Another French psychiatrist, Philippe Pinel, removed the chains from patients and thus initiated the program of scientific, humane treatment towards individuals afflicted with mental aberrations. He divided the field of mental illness into mania, melancholia, dementia, and idiocy. In the last area, research has moved slowly but the present interest and concern over these mentally defective children, provided a new impetus which has ignited vast fields of research. Karl Shapiro (56), a contemporary poet, voices the layman's attitude towards mental deficiency in his poem Mongolian Idiot. He paints a pathetic picture of mongolism, which the modern scientist is endeavouring to correct. (See Appendix I)

Advances in biochemistry, physiology and more refined methods in genetics, has shown that mental deficiency may be due to a malfunctioning of parts of the body; the insulting agent being either endogenous or exogenous. The removal or correction of the disturbing influence may, and in some cases, has allowed the child to progress at a normal rate of development. Selected clinical disorders have provided encouraging results in this new approach. A dramatic example may be seen in phenylketonuria; a disorder where an enzyme system is inadequate to convert phenylalanine to its normal metabolite,
tyrosine.

The main group of mental defectives chosen for this study was the mongoloids. Although their etiology has been discussed extensively elsewhere (1, 2, 3, 4, 10, 11, 15, 20, 21, 27, 32, 33, 34, 35, 36, 37, 39, 44, 45, 50, 52, 54, 57, 58, 60) suffice it to say here that the disorder is primarily one of prenatal dysfunction. That the maturational processes have been arrested at this stage is indicated clearly through the physical malformation and mental development they display. As to the causation of the anomaly, considerable doubt exists whether it is of constitutional or environmental origin. Leader (1932) noted that when the general development of a person is arrested a pathological development of the vessels results. Instead of the capillaries developing in a normal way they tend to retain their primitive shapes and to assume bizarre forms. This forms the basis for the selection of mongolism to study. If Leader's statement is correct, the mongoloid group should display primitive and bizarre morphology.

I. THE PROBLEM

Statement of the problem. The purpose of the study was to test the hypothesis (1) that the mental defectives as a group differed from the normals as a group in their capillary morphology; (2) that differences exist within the defectives in relation to their capillary structure and, (3) that the capillary morphology may be used as a diagnostic tool for the border-
Importance of the study. The primary aim of the present study was to consider the group of mongoloids in order to establish another physical correlate which may be instrumental in assisting the clinician in his diagnosis. The ability to diagnose the borderline cases is of importance. It is desired that the material collected will increase the data that is accumulating concerning this disorder. It is hoped that in the not too distant future all the relevant facts may be drawn together and a solution found to the problem. A quotation from Harley Williams' *Don Quixote of the Microscope* (63), is apropos at this point, "Ramon y Cajal had found the brain an impenetrable forest, but had turned it into a delightful park".

Definition of patterns. The classification the present study is compared with found that the "normal" distribution of capillaries was a series of discrete categories. These have been defined by the authors (Gibson, Bosley & Griffiths 1956) and are reproduced below.

**Pattern A** - This is represented by those capillaries that have no warpings or indentations of either the afferent or the efferent limb, no twisting of either limb about itself or its opposite component, and no excessive distention of either the afferent or the efferent limb, or the tip of the loop.

**Pattern B** - This is represented by those capillaries that show a certain amount of warping or indentation of one or both limbs. No twisting of one limb around the other or itself is evident.
Pattern C - This is represented by those capillaries that show a single crossing in their passage to and from the sub-papillary plexus of deeper vessels. They may also show some warping or indentation, but the single twist is the prime consideration in classification.

Pattern D - This is represented by those capillary loops whose limbs show more than one twist about each other. They may, in addition, show some warplings, indentations or excessive distention.

Pattern E - This is represented by those vessels in which the main feature is an obscure twisting of one limb upon itself. This may be seen in either or both of the arms. Sometimes a certain amount of twisting similar to the type seen in Pattern C is evident also.

Pattern F - This is represented by those capillaries with an anomalous morphology at the tip of the loop. The general picture resembles the head of a femur in outline. This anomalous morphology usually extends into the efferent limb.

Pattern G - This is represented by those capillaries that do not clearly spring from the deeper sub-papillary plexus. They appear to represent an immature horizontal network of vessels.

Pattern H - This is represented by capillaries that resemble flowery shapes with many diverse twistings. They are usually indistinct in outline, presenting a typical blurred appearance.

Pattern I - This pattern is represented by an extreme modification of Pattern B. The loops present a meandering course as they pass from the afferent to the efferent limb. A horizontal plexus is sometimes plainly visible.

Pattern J - This pattern is represented by very short, thin vessels.
CHAPTER II

REVIEW OF THE LITERATURE ON CAPILLAROSCOPY

As early as 1663, Johan Christophorus Kolbasia explored capillary microscopy as a new tool which could be used as a possible diagnostic procedure. His success in this venture is unknown, but he laid the cornerstone which was to have great importance.

In 1866 Stricker, following his studies on the excised membrane of the frog, described the structure and function of capillaries. Rouget in 1873 reported an independent action of the capillaries which was brought about by means of spindle shaped, smooth muscle cells which had the power to contract and expand. In 1893, Spalteholz reported his careful anatomic studies of the vascular system of the human skin which he found to be supplied from the underlying tissues through a large number of small arteries forming an anastomosing irregular plexus. Heimberger made an observation of considerable interest. He found that around each capillary loop in the papillae there is a continuous lymphatic sac, traversed by a number of threads or septums, which hold the capillary loop in place.

It was not until 1912 that Lombard carried out the pioneer work in the field. He found that if a drop of glycerine was placed on the back of the hand previously cleaned with alcohol and a bright light brought to bear on the area, a beautiful picture of the papillae and the superficial vessels
was obtained. By choice the area examined became the fingernail bed, for here the capillaries are arranged in a horizontal plane and generally only one layer thick. Many experimenters have worked in this area to show capillary morphology by photomicrographic means (6, 12, 14, 19, 24, 25, 26, 28, 29, 30, 62, 64).

Since the use of capillary microscopy began, all the early studies by both American and German authors showed that there is a definite and direct relationship between the development of the capillary structure and mental development. Examinations of German authors (reported by Hauptman and Myerson, 1948) show that the capillaries of school children have a normal pattern in 67% of the cases, whereas children observed from an ungraded school had normal capillaries in only 29% of the cases. The authors suggest that capillaroscopic examination may help in the differential diagnosis of mental abnormalities arising either through constitutional or environmental etiology.

Powdermaker (1929) made one of the earliest attempts to relate structure to function. She studied the capillary form in relation to constitutional and secondary hypophrenias. The importance of this study lies in the possibility of assisting in the early detection of a constitutional defect and in differentiating it from an environmental defect. Of particular note in Powdermaker's study was her observation that in constitutional idiots and low grade imbeciles, in whom there was a high proportion of physical abnormalities (88%), there was a
marked tendency for a retarded or abnormal development of the capillaries; this was much less marked in the few cases which showed in approximately normal physical development. In the moron and high grade imbecile group there were relatively few primitive forms. In the cases in which these forms were found, there was a significantly greater proportion of emotional, endocrine and neurologic disorders. No primitive forms were found in cases of secondary origin and in stable children with normal, physical and mental development.

Jaensch, with Hoepfner and Wittneben, in the same year, published a book from a study of 3000 cases. They asserted that when the general development is markedly defective the capillaries may never lose the main characteristics of the primitive network form, and both limbs of the capillary loops will have a wide calibre.

Morphology of the normal capillary pattern. The problem of determining a normal form of capillaries is as old as the first observation. There is considerable discrepancy throughout the literature concerning this matter, but most of the data can be assigned to one of two classifications. The first category designates the normal pattern, for the nail bed as consisting of straight forward hairpin loops. In this group, any deviations from the "normal" form constitutes relative "abnormalities" (8, 9, 38, 41, 42, 46, 51, 53, 59). The other category is designated by more than one form as consisting of
the "normal" pattern. The authors in this group have concluded that it is difficult to establish a "normal" pattern due to the wide variations found in the control groups (7, 12, 13, 16, 17, 23, 24, 25, 26, 28, 29, 30, 40, 41, 42, 43, 55, 64).

Gibson, Bosley and Griffiths (1956), recognizing the importance of the problem, set out to determine a normal morphological pattern. They selected four hundred "normal" individuals and capillaroscopic examinations were recorded. An analysis of the results revealed not one but several forms were indicative of a normal pattern. The chief limitation of adopting this procedure lies in statistical analysis. The data collected indicates capillary morphology on a total basis only and does not provide the morphology for a single nail bed. In the present study, a comparison between clinical groups was desired, therefore, this classification was used.

Powdermaker (1929) has classified the vessels based on degrees of maturity, with the final stage (straight hairpin loop) representing the "normal" pattern. Since her classification is based on the maturation of the vessels, this is an opportune time to discuss the development of the capillaries.

The developmental sequence begins with an arabesque of vessels with no particular order. It is an anastomosing arteriolar plexus arising in the deepest layer of the dermis present at birth. There are no true capillary loops and the upper end of the corium represents a flat line. At one month the subpapillary plexus begins to disappear and up to four or five
months saddle shaped forms sprout from the primitive plexus. They grow gradually in the direction of the corium; but not before the fifth month does one find a few hairpin-like capillary loops with differentiation in calibre. At the same time, the horizontal network of fine blood vessels increases in size and complexity. The corium becomes wavy and assumes the characteristic scalloped appearance with each capillary growing into one papillae. From the fifth or sixth month to one year the capillaries develop to the mature picture with a disappearance of the wide loops and subpapillary plexus.

At this particular stage the capillary morphology is considered complete. The pattern is persistent, changing only with old age, and any tortuosity in children is considered abnormal. (Leader, 1932).
CHAPTER III

EXPERIMENTAL METHOD

The problem as stated necessitates a capillary assessment on two groups of mental defectives. The capillary analysis was determined after the classification of Gibson, Bosley and Griffiths (1956). A further analysis was made on whether a subpapillary plexus was present, for Hauptman and Myerson (1948) stress that in normal individuals, no subpapillary plexus is seen.

Subjects. The experimental groups for this study consisted of one hundred and twenty-six mongoloids and sixteen phenylketonurics taken from Woodlands School for Mentally Retarded Children, New Westminster, B.C. Mongolism was chosen because it is one of the largest groups of mental defectives. The group phenylketonuria was selected because of the interest it has aroused since Polling first noted the disorder in 1934 as one of the relatively few "inborn error" of metabolism. Although the total population of the two groups were used, the thickness of the epithelium prevented obtaining photomicrographs in every case. This, however, is a technical difficulty that was unavoidable.

Method. The equipment used folded very conveniently into two carrying cases. Because of the relative ease to move the
equipment, the patients were examined on the wards at the school. The fourth finger, left hand, was selected for observation. It was prepared with zylene, dried, and cedar oil then spread over the nail bed.

**Apparatus.** A modified Zeiss Opton standard stereoscopic binocular dissection microscope was used. An extra arm was fitted for the finger clamp assembly. A 35 mm. Canon camera with a Mecablitz electronic flash was attached. The electronic flash (1/500 second) proved an invaluable addition since it removed the difficulty of aperture settings and moving fingers. An auxiliary Leitz Wetzlar skin and capillary microscope lamp was used for focusing. (See Figure 1).

The prints were made on Kodak, Linagraph Ortho safety film; a very fast orthochromatic film of moderate contrast. The film was developed in Kodak D 19 and enlargements made on Kodabromide paper. It was found that viewing the film strips in a microfilm reader was most satisfactory. The pictures could be enlarged, focused and the intensity altered instantaneously. A Tri-purpose projector, manufactured by the Society for Visual Education, Inc., with a Powerstat, manufactured by the Superior Electric Co. were used in projecting and illuminating the film. (See Figure 2).
Figure 1: The capillary microscope with attachments.
Figure 2: The microfilm reader with reostat used for viewing the capillary film strips.
A total of one hundred and twenty-six mongoloids were used for the determination of capillary morphology. These results were contrasted against sixteen phenylketonurics, and both mentally defective groups were compared with the normal group of Gibson, Bosley and Griffiths (1956).

The above mentioned investigators formulated a basic capillary pattern that appeared to be common to the majority of "normal" healthy individuals. This pattern consisted of the following forms designated A to J respectively.

1) Hypothesis: The group of mongoloids display a morphological pattern no different from that found in the normal population. (See Figure 3).

Computed chi square of 109.1 was significant at the 0.01 level of confidence.

Therefore the hypothesis is rejected.

2) Hypothesis: The group of phenylketonurics display a morphology similar to the normal population (See Figure 4).

Computed chi square of 296.20 was significant at the 0.01 level of confidence.

Therefore the hypothesis is rejected.
Basic morphological pattern of mongoloids compared to normal group of Gibson, Boasley and Griffiths (1956).
Figure 4
Basic morphological pattern of phenylketonurics compared to normal group of Gibson, Bosley and Griffiths (1956).
3) Hypothesis: There is no difference in morphological pattern within the mentally defective groups of mongolism and phenylketonuria (See Figure 5).

Computed chi square value of 23.41 was significant at the 0.01 level of confidence.

Therefore the hypothesis is rejected.
Figure 5

Basic morphological pattern of mongolism and phenylketonuria.
The primary aim of the present study was to determine the morphological pattern of the capillary structure of the mongolian idiots (See Figure 6). The resulting pattern is presented in Figure 3, along with a comparison to a "normal" group. It was observed that the mongoloids differed significantly from the "normal" population.

The number of open loops (A and B) were reduced in the mongoloids. As has been previously stated, the type A structure is considered by many to be representative of the normal pattern. In type G the difference is enormous since this pattern is seldom found. Pattern G has been described as those capillaries which represent an immature horizontal network of vessels; thus with the increased appearance of Patterns G, H and I, the mongoloid group displays a rather immature and undeveloped pattern (See Figure 7). As observed previously, mongolism is the result of foetal arrest, thus there will be an increased number of primitive forms since the delay in capillary development should be correlated with the general structural or physical retardation.

It has been stated by many authors (29, 30, 38, 53,) that the presence of a subpapillary plexus Is indicative of an immature configuration. Gibson, Bosley and Griffiths (1956) did not report the presence or absence of a plexus in their normal
groups studied, therefore, reference to other literature on the subject has been resorted to, in order to support the discussion. It is interesting to note that within the mongoloids the plexus occurred only in the early age levels. There were no cases past the age thirteen in the females (which comprised 63.4% of the cases) and age eighteen (which comprised 68.4% of the cases) in the males. This implies that in the remaining cases the configuration has progressed beyond the plexus to a more mature stage. If this implication is correct, then a very interesting inference can be made.

Hauptman and Myerson (1948) observed that in mental retardation which has as its origin an endogenous agent, the capillary structure will be immature and will never undergo development at later periods. When this is interpreted in light of the above observation, it becomes of paramount importance to the age-long query whether the syndrome mongolism has its etiology in constitutional or environmental factors. If the precipitating cause of mongolism is constitutional, then the capillary pattern should be primitive and never mature. However, if the agent operating is exogenous, that is, operating through the maternal environment, then the pattern may alter with age, losing some of its undeveloped structure. Since this appears to be the case it would seem that the etiology of mongolism exists in environmental and not solely constitutional factors.

Mongolism was compared with another group of mental
defectives the phenylketonurics. It was desirable to know whether mongolism differed from other mental deficiencies, since this information is necessary before the morphological pattern evolved could be used as an aid diagnostically. It was noted that the mongoloids differed significantly from the phenylketonurics. Both groups displayed a high frequency of plexus (See Figure 8). In the mongoloid group there was 23.1% while in the phenylketonuric group 56.2% occurred.

Powdermaker (1929) noted that a correlation exists between the poorly developed capillaries and a general defect in the development of what has been called the psychophysical personality. Leader (1932) noted that when the general development of a person is arrested a pathologic development of vessels results. Instead of the capillaries developing they tend to retain their primitive shapes and assume bizarre forms.

Following the observation that the mongoloids and phenylketonurics retained many primitive forms (See Figure 9), especially the plexus, it was thought advisable to examine a group of children who were intellectually retarded so that a comparison with a less severely retarded group could be viewed in respect to a correlation existing between the resulting behavior and the physical status.

Normally, it is considered by psychologists that intelligence is primarily inherited and consequently of constitutional origin (Munn 1951). The constitutionally retarded individuals whose malfunctioning was severe (mongoloids and phenylketonurics)
Figure 6: Photomicrograph of the capillary nailbed of a mongoloid displaying the typical pattern of an underlying plexus with a tortuous morphology.
Figure 7: Photomicrograph of the capillary nailbed of a mongoloid showing an immature plexus with corresponding undeveloped morphology.
Figure 8: Photomicrograph of the capillary nailbed of a phenylketonuric showing plexus and undeveloped morphology.
Figure 9: Photomicrograph of the capillary nailbed of a phenylketonuric displaying an immature morphological pattern.
displayed a large proportion of undeveloped morphology (See Figure 10) which appeared to correspond with the less severely retarded individuals (Dawson Summer School students). Of the thirty-six students a total of 22.2% possessed a plexus and retained much of the undeveloped structure as evidenced by the increased occurrence of pattern G and I (See Figure 11). Thus it appears that Powdermaker's and Leader's observations of a correlation existing between structure and function is tenable. Therefore, it seems that the undeveloped pattern of the mental defectives bears a relationship to their psychopathology and the presence of a plexus is indicative of retarded development. This conclusion is upheld by the similarity with the less retarded students.

1. Operated by the Y.M.C.A. for children in grade school who have to repeat their year or have considerable difficulty with their school program.
Figure 10: Photomicrograph of the capillary nailbed of a student at Dawson Summer School showing the typical immature morphological pattern with plexus.
Comparison of basic morphological pattern of retarded school children with mongoloids and phenylketonurics.
CHAPTER V

SUMMARY

A capillaroscopic examination of the clinical groups, mongolism and phenylketonuria, was carried out at the Woodland's School for Retarded Children.

The capillaries were analysed after the ten basic morphological patterns set out by Gibson, Bosley and Griffiths (1956).

A comparison was made within constitutional retardation of severe (mongolism and phenylketonuria) and less severe (Dawson Summer School students) order.

CONCLUSIONS

1. The mongoloids and phenylketonurics, as a group, differed significantly from normals, as a group, in their capillary morphology.

2. There was a significant difference found between the morphological pattern of mongoloids and the phenylketonurics.

3. Capillary morphology may be used on a group basis to assist in diagnosis of borderline cases of mongolism.

4. There was a similarity between the morphological patterns of severe and less severe constitutionally retarded individuals.
REFERENCES


APPENDIX I

Mongolian Idiot

A dog that spoke, a monster born of sheep
We mercilessly kill, and kill the thought,
Yet house the parrot and let the centaur go,
These being to their nature and those not.
We laugh at apes, that never quite succeed
   At eating soup or wearing hats.

Adam had named so many but not this,
This that would name a curse when it had come,
Unfinished man, or witch, or myth, or sin,
Not ever father and never quite a son.
Ape had outstripped him, dog and darling lamb
   And all the kindergarten beasts.

Enter the bare room of his mind and court
His store of words with letters large
See how he handles clumsily those blocks
With swans and sums; his colored picture books.
At thirty-five he squeals to see the ball
   Bounce in the air and roll away.

Pity and fear we give this innocent
Who maimed his mother's beautiful instinct;
But she would say, "My body had a dog;
I bore the ape and nursed the crying sheep.
He is my kindness and my splendid gift
   Come from all life and for all life."