

EFFECTS OF INTRAUTERINE RUBELLA INFECTION AND ITS CONSEQUENT PHYSICAL SYMPTOMS ON INTELLECTUAL ABILITIES

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The assumption commonly made that rubella infection produces mental retardation was evaluated psychometrically using an instrument independent of the frequently deficient hearing and speech abilities of individuals so affected. Eighty-eight testable children with evidence of rubella infection earned a mean IQ score of 100.01 on the Arthur adaptation of the Leiter International Performance Scale. It was discovered that the number of physical symptoms found was mildly related to intellectual level ($r = -.21$, $p < .025$). A group of untestable blind-deaf children was also examined briefly, and the overall conclusion was drawn that it is not the rubella infection per se that produces the clinical picture commonly seen as one of retardation but rather the physical symptoms that limit psychological development and expression.

Of current concern to the public health field are the multiple congenital defects caused by the intrauterine infection of the rubella virus (German measles). Epidemics of the disease tend to occur every 6-9 years (Lin-Fu, 1970), with the most recent 1964-1965 epidemic resulting in an estimated 30,000 children in the United States with birth defects (Cooper, 1968). With another epidemic expected early in this decade, efforts are being made nationally to vaccinate potential victims and carriers, and this preventive measure may prove to be the ultimate answer.

Presently, however, evidence abounds for the physical defects from which children suffer. The defects most commonly reported include auditory impairment, visual irregularities (cataracts, retinopathy, amblyopia, myopia), and congenital heart disease, although a large number of additional problems have been reported in infected individuals, including various physical malformations, asthma, migraine, neonatal thrombocytopenic purpura, genital defects, etc. (Cooper, 1968; Hardy, McCracken, Gilkeson, & Sever, 1969; Karmody, 1968; Miller, Rabinowitz, Frost, & Seager, 1969; Sheridan, 1964). The prevalence of the more common anomalies is indi-

cated by Cooper (1968), who in a study of 271 rubella-infected infants found that 52% had hearing losses confirmed or suspected, 40% had cataracts or glaucoma, and 52% had congenital heart disease. The close correspondence of clinical problems with the presence of the rubella antibody which indicates congenital infection is pointed to by this study since only 9 of the 500 children studied were considered clinically *normal*.

With so many physical defects present, and especially those that affect the development of intellectual functions such as vision and hearing, it is not surprising to find either direct statements or inferences of retardation associated with the syndrome (e.g., Coleman, 1972; Cooper, 1968; Heiser & Wolman, 1965; Lin-Fu, 1970; London & Rosenhan, 1968; Menser, Dods, & Harley, 1967; Plotkin, Cochran, Lindquist, Cochran, Schaffer, Scheie, & Furukawa, 1967; Ullmann & Krasner, 1969). Some of these investigators have clinically estimated up to one third of their populations to be retarded. The assumption seems to be made that both the physical problems and the seeming retardation are physically caused by the viral infection. However, no systematic effort has been made to see if these individuals are in fact retarded in testing situations where these children are not handicapped in their communication by their speech and hearing problems.

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Considerable literature exists in the general area of physical handicaps and intelligence, and as it has already been reviewed extensively (e.g., Dinnage, 1972; Wolf & Anderson, 1969), no effort will be made to do so here. However, it is worth noting that a number of studies (e.g., Greenbaum & Buehler, 1960; Ross, 1972) point to relationships between these factors with populations other than rubella-infected children, and hence the possible effects of such must be taken into account.

Sheridan (1964) in the one extensive psychometric effort with rubella-infected children made to date, was able to obtain intelligence test scores on 191 English children with evidence of rubella infection. The basic finding was that the mean IQ score was slightly above average (106.8), and that the distribution approximated normality. However, a number of factors prevent these results from being conclusive, including the facts that data were not available on the complete rubella sample under study, testing results were accepted by mail from a variety of sources without knowledge of standard conditions of administration, IQ scores were combined from several tests indiscriminately, and there was a lack of attention to test results that may have been invalidated by communicational deficiencies. However, the conflict between the general results of the study and clinical observation as typically reported points to the need for either confirming or disconfirming the findings.

From the above, it is clear that while most investigators seem to almost automatically assume that rubella physically results in retardation, it is not clear either that rubella-infected children are retarded in any general way, or that any such retardation is due to the infection itself rather than as secondary effects of the physical symptoms. This study addresses itself to this basic question and, in addition, asks the question of whether there is any relationship between the number of physical anomalies seen and intellectual level as assessed.

METHOD

Subjects

The Crippled Children's Division of the University of Oregon Medical School served as a means by

which a reasonably representative subject population could be gathered. This state-supported agency provides services for handicapped children across the state, and while utilized by only a portion of the eligible individuals in the state, it services persons from all socioeconomic levels. All children known to the clinic who were born in 1964, 1965, or 1966 and who were suspected as possibly suffering from rubella infection were examined in detail for this study.

The criteria for acceptance into the study as having contracted rubella were either a positive titer between 3 and 24 months of age (a positive culture of the rubella virus in the infant's blood—Plotkin et al., 1967) or a history of rubella or or rubellalike illness during gestation *plus* the presence of (a) congenital heart disease, (b) a congenital eye disorder (cataract, glaucoma, or retinopathy), or (c) any degree of impairment in hearing not conductive in nature. The symptoms were selected because they have clinically been shown to appear following rubella infection by a number of investigations (Coleman, 1972; Cooper, 1968; Plotkin et al., 1967). Alone, they would not necessarily indicate rubella, but in combination with a history of rubella or rubellalike infection and medical examination revealing no other cause of the physical disorders, evidence for rubella infection was accepted.

In setting up these multiple criteria, it was our purpose to ensure that those individuals included were definitely affected by the rubella virus, even though in so doing it was possible that an occasionally mildly infected individual was not identified. Hence, if the population is biased, it is biased in the direction of identifying the more severely affected children.

A total of 109 children met the criteria as being infected with the rubella virus. Of these, 16 had significant impairment in both hearing and sight so that they were untestable. Severe behavior problems existed in 3 subjects so that they could not be properly examined, and incomplete data existed on 2 others. The remaining 88 subjects (39 males and 49 females) were used as the principal subject pool, and their mean age at the time of testing was 57.00 months ($SD = 9.18$ months).

Procedure

All children received thorough medical, audiologic, and psychological examinations followed by a team staffing. The data used in the present study were obtained from the comprehensive evaluation. The Arthur adaptation of the Leiter International Performance Scale (Arthur, 1949) was selected as the basic instrument for intellectual evaluation. Since this instrument employs no speech at all but only the manipulation of blocks into slots according to the principles being tested, it was suitable for the evaluation of this type of population, a substantial portion of which had marked hearing and speech problems.

TABLE 1
FREQUENCIES OF PHYSICAL SYMPTOMS IN 88 RUBELLA-
INFECTED CHILDREN

Symptom	Frequency
Congenital heart disease	42
Nonconductive hearing impairment	81
Congenital eye disorders	
Glaucoma or cataract	13
Retinopathy	72
Short stature	36
"Extended" syndrome	7
Encephalopathy	9

Note should be made that the Arthur adaptation of the Leiter scale avoids the standardization problems associated with the original sample (Leiter, 1940) and produces results more nearly in line with other examinations in the area such as the Stanford-Binet and the Wechsler Intelligence Scale for Children (WISC) (Tate, 1952). In using the instrument in this study, the contention is not that the instrument is the best possible assessor of intelligence, but rather that it is a better predictor with the handicapped population in question than either the language-dependent Binet or the WISC. Comparisons between the Arthur revision and these other instruments may be cited (Alper, 1958; Cooper, 1958; Orgel & Dreger, 1955; Tate, 1952; Werner, 1965), with the overall conclusion being that the Arthur revision slightly underestimates the intelligence scores rendered by other instruments.

The major and minor manifestations of the rubella syndrome were documented by medical and audiological team members. All subjects had formal ophthalmological examinations, and the difficulties discovered were subclassified into cataract or glaucoma and retinopathy. Cardiologic, orthopedic, and neurologic consultations were obtained where indicated. In addition, special attention was paid to three less specific manifestations of the rubella syndrome. These were shortness in stature (greater than one standard deviation below the mean compared with children their age), the "extended" syndrome (existence of purpura, jaundice, enlarged liver and spleen in the newborn period), and encephalopathy. Hence, each person was evaluated on the presence or absence of seven types of physical symptoms, and the number of physical symptoms present for each was noted. Table 1 gives summary information with respect to the presence of all physical symptoms.

Analysis

The mean and standard deviation of the Leiter scores were computed and examined to assess the null hypothesis of no difference between rubella and normal children. Also, the question of whether those scores were related to the number of physical symptoms was assessed by correlational procedures

because both the literature (e.g., Greenbaum & Buehler, 1960; Ross, 1972), and clinical observation suggested that an inverse relationship between these variables might be found. Finally, a preliminary effort was made at inferring the intellectual level of the group of deaf-blind children from the physical symptom data.

RESULTS

The 88 children had a mean IQ of 100.01, with a standard deviation of 18.94 and a range of 52-153. Figure 1 shows the distribution of scores to be essentially normal, especially on the side of the distribution of interest relative to the question of retardation. Hence, the pattern of scores found is generally that expected with a normal group of children.

The mean number of physical symptoms present in the group was 2.95, with a standard deviation of 1.20 and a range of 1-6. The correlation between the number of physical symptoms present and IQ was $-.21$, which was significant ($p < .025$, one-tailed t statistic). Hence, there was a mild but dependable relationship between these variables.

DISCUSSION

The results on the general intellectual level of the rubella-infected children who were testable is beyond question: they are clearly normal in intelligence. Hence, in contrast with general inferences frequently made, one

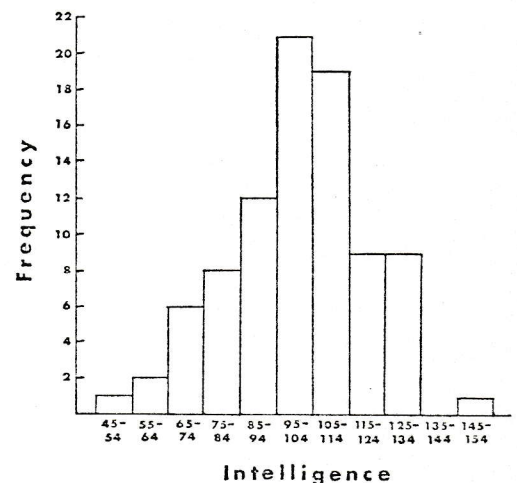


FIGURE 1. Distribution of intelligence test scores for a group of 88 rubella-infected children.

might conclude that rubella had no direct limiting effect on the intellectual potential of these children.

However, there is a constraining factor here, and it relates to the group of 16 children who were both deaf and blind and so could not be tested. Many of them were clearly functioning at a low level, whatever the cause. One might hypothesize that here are retarded individuals who if they could be tested would make the distribution of scores an abnormal one. This is an exceedingly difficult question to assess, but a very tentative means of exploring it was possible through the data on physical symptoms that were available for 15 of these children. If one gives each of these children the mean IQ score of the part of the larger group having the same number of physical symptoms as he has, then a series of 15 scores will result. The mean of these scores is 98.29. Again, this is of the most speculative nature, for it rests on the assumption that all symptoms are equal, or at least that differences between them would cancel out. What is surprising about the score of 98.29 is that it is not lower.

Could it be that the deaf-blind children would approximate this level if they were not blind (i.e., if they were testable)? Admittedly, they do have more physical symptoms ($\bar{X} = 3.40$; $SD = .40$) than the larger group ($t = 2.30$, $p < .05$, two-tailed test), but while such symptoms are reliably related to intelligence, only about 4% of the variance in intelligence scores can be related to the symptoms. Is there any reason to believe that an additional symptom (blindness) should necessarily result in a loss of intellectual potential? No, but yet it is this one symptom that differentiates the testable from the nontestable groups. Therefore, it was hypothesized that it must not be the physical damage from rubella that results in functioning at the level of retardation, but rather the secondary effects of such damage, namely a cutting off of information to the brain. An attempt was made to substantiate this further by subdividing the symptoms into those that were directly related to cognitive development (hearing impairment, glaucoma or cataract, encephalopathy) and those that were not (heart disease, retinopathy, short stature,

"extended" syndrome) and then to relate frequencies in these two subdivisions to intelligence. However, a severe restriction in range resulted in the number of symptoms in each category, and appropriate analyses therefore could not be conducted.

In conclusion, this study has shown that children with a history of intrauterine rubella infection who have the important sensory modality of vision remaining are not retarded, but rather have average intelligence. Physical symptoms seem important primarily as they close off important sources of information to the developing child. The suggestion, then, is that the clinical picture commonly seen as one of retardation is *not* produced by the physical damage of the rubella virus per se, but rather by the secondary effects of such damage that limit psychological development and expression. If this is true, then we may repudiate the notion that rubella results in inevitable mental retardation, and we may move on to more optimistic and more realistic treatment approaches.

REFERENCES

- ALPER, A. E. A comparison of the WISC and the Arthur adaptation of the LIPS with mental defectives. *American Journal of Mental Deficiency*, 1958, 63, 312-316.
- ARTHUR, G. The Arthur adaptation of the Leiter International Performance Scale. *Journal of Clinical Psychology*, 1949, 5, 345-349.
- COLEMAN, J. C. *Abnormal psychology and modern life*. (4th ed.) Glenview, Ill.: Scott, Foresman, 1972.
- COOPER, J. G. Predicting school achievement for bilingual pupils. *Journal of Educational Psychology*, 1958, 49, 31-36.
- COOPER, L. Z. Rubella: A preventable cause of birth defects. In D. Bergsma (Ed.), *Birth defects: Original article series*. Vol. 4. *Intrauterine infections*. New York: The National Foundation, 1968.
- DINNAGE, R. *The handicapped child: A research review*. Vol. 2. London: Clowes, 1972.
- GREENBAUM, M., & BUEHLER, J. A. Further findings on the intelligence of children with cerebral palsy. *American Journal of Mental Deficiency*, 1960, 65, 261-264.
- HARDY, J. B., MCCracken, G. H., JR., GILKESON, M. R., & SEVER, J. L. Adverse fetal outcome following maternal rubella after the first trimester of pregnancy. *Journal of the American Medical Association*, 1969, 207, 2414-2420.
- HEISER, K. F., & WOLMAN, B. B. Mental deficiencies. In B. B. Wolman (Ed.), *Handbook of clinical psychology*. New York: McGraw-Hill, 1965.

- KARMODY, C. S. Subclinical maternal rubella and congenital deafness. *New England Journal of Medicine*, 1968, 278, 809-814.
- LEITER, R. G. *The Leiter International Performance Scale*. Santa Barbara: Santa Barbara State College Press, 1940.
- LIN-FU, J. S. *Rubella*. Washington, D.C.: U.S. Department of Health, Education, and Welfare, 1970.
- LONDON, P., & ROSENHAN, D. (Eds.) *Foundations of abnormal behavior*. New York: Holt, Rinehart & Winston, 1968.
- MENSER, A., DODS, L., & HARLEY, J. D. A twenty-five-year follow-up of congenital rubella. *Lancet*, 1967, 2, 1347-1350.
- MILLER, H., RABINOWITZ, M., FROST, J. O., & SEAGER, G. M. Audiological problems associated with maternal rubella. *Laryngoscope*, 1969, 79, 417-426.
- ORGEL, A. R., & DREGER, R. M. A comparative study of the Arthur-Leiter and Stanford-Binet intelligence scales. *Journal of Genetic Psychology*, 1955, 86, 359-365.
- PLOTKIN, S. A., COCHRAN, W., LINDQUIST, J. M., COCHRAN, G. C., SCHAEFFER, D. R., SCHEIE, H. G., & FURUKAWA, T. Congenital rubella syndrome in late infancy. *Journal of American Medical Association*, 1967, 200, 105-111.
- ROSS, R. T. Behavioral correlates of levels of intelligence. *American Journal of Mental Deficiency*, 1972, 76, 545-549.
- SHERIDAN, D. Final report of a prospective study of children whose mothers had rubella in early pregnancy. *British Medical Journal*, 1964, 2, 536-539.
- TATE, M. The influence of cultural factors on the Leiter International Performance Scale. *Journal of Abnormal and Social Psychology*, 1952, 47, 497-501.
- ULLMANN, L. P., & KRASNER, L. *A psychological approach to abnormal behavior*. Englewood Cliffs, N.J.: Prentice-Hall, 1969.
- WERNER, E. E. Review of the Leiter International Performance Scale. In O. K. Buros (Ed.), *The sixth mental measurements yearbook*. Highland Park, N.J.: Gryphon Press, 1965.
- WOLF, J. M., & ANDERSON, R. M. *The multiply handicapped child*. Springfield, Ill.: Charles C Thomas, 1969.

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