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DOWN SYNDROME: THE BENEFIT OF A HOME PROGRAMME FOR CHILDREN WITH DOWN SYNDROME AS MEASURED ON THE GRIFFITHS DEVELOPMENT SCALES

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DOWN SYNDROME: THE BENEFIT OF A HOME PROGRAMME FOR CHILDREN WITH DOWN SYNDROME AS MEASURED ON THE GRIFFITHS DEVELOPMENTAL SCALES

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A RESEARCH REPORT SUBMITTED TO THE FACULTY OF MEDICINE, UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG IN PARTIAL FULFILMENT OF THE REQUIREMENTS FOR THE DEGREE OF MASTER OF SCIENCE IN DEVELOPMENTAL PAEDIATRICS

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ABSTRACT

This study presents the developmental changes in 20 children with Down Syndrome after participating in an early intervention programme (START) for a minimum period of six months, using the Griffiths Development Scales as outcome measures. It is a descriptive ex post facto study comparing developmental change (DQ) in those children with Down Syndrome who participated in the intervention programme with a non-participating or control group matched for age, sex and socio-economic status. All children with Down Syndrome were diagnosed clinically and serologically. The data was collected over ten sessions, and included a formal child assessment, using Griffiths Development Scales, parent or caregivers interviews and questionnaires. Analysis indicated that the START programme was associated with a statistically significant improvement in the domain of speech and hearing, and also with the onset of developmental milestones such as sitting, walking and speech. However, there was no statistically significant difference in the developmental quotient (DQ) of the two groups. The results were critically examined in terms of methodological issues around acquisition of a pure control group citing the ethical imperatives to provide treatment, leading to difficulties in sampling for age and degree of impairment. From the results it was concluded that the beneficial effects may have been due to caregiver variations although the START programme may still have had a role to play.

DEDICATION

This work is dedicated to my husband Lehlohonolo, my children Ntsane, Tseliso and Mpho and my mother Notozi.

DECLARATION

I, Irene B. Nosiba Skenjana, hereby declare that this research report is my own unaided work and has not been prescribed for any degree of another University.

SIGNED AT ..... ON THIS ..... DAY OF ....., 1999.

.....

IRENE B. NOSIBA SKENJANA

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8. All the children who participated in this study, as well as their parents and caregivers.

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CHAPTER ONE : INTRODUCTION

1.0 OVERVIEW

In this chapter a general overview of Down Syndrome will be discussed, as well as the improvement in the survival rate of people with Down Syndrome over the past few decades. Subsequently , the evolution of attitudes of doctors in the management and approach to Down Syndrome in the light of the changes in the demography of the condition, will be reviewed. Finally, the place of a primary physician/paediatrician in the management of people with Down Syndrome will be highlighted.

1.1 BACKGROUND

Down Syndrome was first described by an English physician, Langdon Down, 1866, in individuals who suffered from a marked arrest of physical and mental development (as discussed in Fishler & Kock, 1991) which occurred in approximately 1 in 1 000 births (Thapar, Irving, Michael & Michael, 1994). There are more than fifty characteristics associated with Down Syndrome (Table 1) and only a few may be present in any one case, except mental retardation which is found in almost all cases. There is seldom more than one sibling or relation afflicted in a family, but it has been reported that between 5 - 10% of these mothers of children with Down Syndrome had other babies with the condition in their immediate family or kindred group (Cunningham, 1986).

TABLE I. FREQUENCY OF PHYSICAL CHARACTERISTICS OF NEWBORNS WITH DOWN SYNDROME

Characteristics	Frequency(%)
Craniofacial:	
Microcephally	50
Flat occiput	60 - 80
Small ears	95
Redundant skin on nape of neck	80
Upslanting palpebral fissure	70 - 90
Epicanthal folds	50 - 70
Brushfield spots	30 - 80
Flat nasal bridge	60 - 80
Narrow short palate	60 - 90
Protruding tongue	40 - 60
Lower lip vertical creases	50
Limbs:	
Short broad hands	70
Simian lines	40 - 60
Typical dermoglyphia	99
Wide space between first and second toe	50 - 90
Increased sole creases	65
Neurologic:	
Hypotonia	40 - 80

(From Rudolphs Paediatrics - A Rudolph (1982: 295 - 296).

The exact aetiology of Down Syndrome is unknown, despite an abundance of theories and hypotheses in past decades. Lejeune, Günter and Turpin (1954), as cited by Fischler and Kock (1991), made a major contribution to the study of Down Syndrome in the area of cytogenetics and identified some chromosomal aberrations particularly the trisomy of chromosome 21. Trisomy 21 refers to the presence of an extra copy of chromosome 21, which in 95% of affected individuals is the result of a failure of the two chromosomes 21 to separate at meiosis or more rarely at mitosis, thus producing an extra chromosome in the embryo (non-disjunction). This process is more commonly maternal in origin, but can occur in either parent. It is associated with increasing maternal age. In approximately 2% of cases Down Syndrome can be attributed to chromosomal translocation, a process which is associated with higher recurrence rate, but not with increasing maternal age (Thapar et al, 1994). In an even smaller number of affected individuals there is evidence of mosaicism, a condition where only a proportion of cells is trisomic. This occurs as a result of meiotic non-disjunction or mitotic non-disjunction occurring after fertilization. It has been argued that many such subjects showed only partial characteristics of Down Syndrome, clinically and mentally, and have been described as having "near-normal" intelligence (Fishler & Kock, 1991).

The risk of a mother over the age of 30 having a child with Trisomy 21 is 1:100 and increases with advancing maternal age, such that at about 40-49 years of age the risk is about 1:2. This is strongly associated with nondisjunction - see Figure 1. If there is translocation, the risk of a mother having a child with Down Syndrome is 1:3. The

mother of a child with Down Syndrome has a risk of 1:50 of producing another affected child (Thapar et al. 1994).

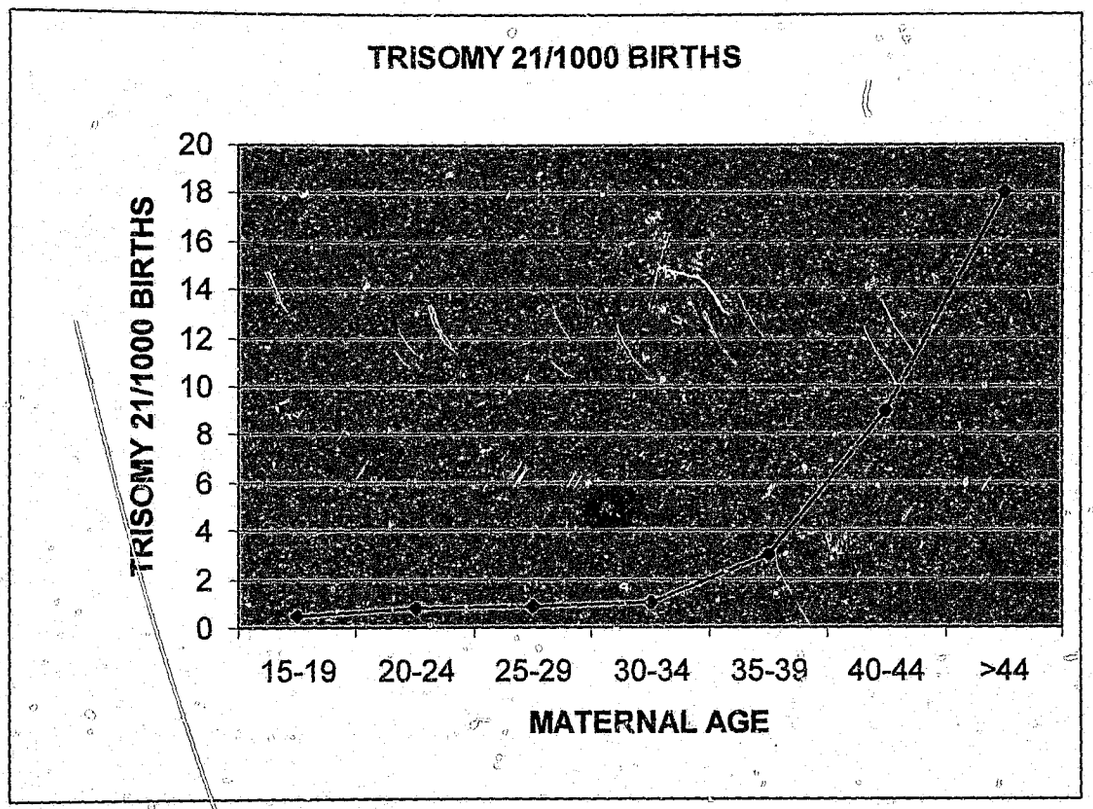


Figure 1: The relation of maternal age to the risk of having a child with Down Syndrome (Modified from C.O. Carter, K.A. Evans, Lancet 2: 785, 1961).

Down Syndrome is recognised as one of the commonest congenital conditions associated with mental retardation in the first world. The incidence of Down Syndrome in the United States of America is 1:700 births and is responsible for 10% of all mental subnormality in children (Thapar, et al, 1994). The British figures are

given as 1:1000 live births. Christianson (1996) contends that numerus attempts to study the incidence of Down Syndrome in sub-Saharan Africa have been inconclusive due to incomplete ascertainment at birth, high mortality in infancy from associated congenital abnormalities and short periods of case collection. He also states that the original data of Kromberg and Christianson (1989) was later refined to 1.8 per 1000 live births for infants born in central Johannesburg and Soweto, and 1.2 per 1000 of the surrounding clinic maternity units were included. Two other studies found the incidence to vary between 1.33 per 1000 live births in a Pretoria urban academic hospital (Delport, Christianson & Gericke, 1995) and 2.09 per 1000 live births in a rural hospital (Venter, Christianson, Humato, Makhura & Gericke, 1995).

Studies world-wide have shown a continuing rise in the life expectancy of persons with Down syndrome, which is largely brought about by an increased survival rate of infants due to, inter alia, improved medical management of the congenital complications associated with Downs Syndrome (Carr, 1993), (Table II.).

TABLE II. PERCENTAGE SURVIVAL RATE OF DOWN SYNDROME CHILDREN BIRTH COHORTS

	UP TO 1 YEAR	UP TO 5 YEARS	UP TO 10 YEARS
PERIOD	**RATE**	**RATE**	**RATE**
1940 - 1950	60%	20%	36,5%
1950 - 1970	80%	71%	69,5%
1976 - 1985	90%	79%	86,5%

Clearly, the life expectancy of people with Down syndrome has increased considerably and the implications thereof are pervasive. The prolonged life span of people with Down Syndrome has had a tremendous impact on research, for example, the understanding of Alzheimer disease in the general population (Carr, 1993). It has also stimulated research into family intervention and outcome, and a better understanding of the expected capabilities, residential placement and work prospects of children with Down Syndrome (Gath, 1990).

1.1.1 TRENDS IN THE CARE OF DOWN SYNDROME

Over the past three decades the treatment of the individual with Down Syndrome and other developmental disabilities has evolved from an era where they were subjected to inadequate custodial care in hospital, where they were left to die with no treatment, to the present era of intervention programmes and improved medical/surgical care.

In the past, the presence of congenital heart disease, poor feeding and mobility skills were important factors in the morbidity and mortality of children with Down Syndrome (Carr, 1993). Medical care was often withheld for example, in 1963, the parents and the attending doctor of an infant with Down Syndrome with duodenal atresia refused corrective surgery and the infant died eleven days later (Gustafson, 1973). Again, the question of life-sustaining treatment for infants with Down Syndrome was raised with the publication of the famous case of "Baby Doe" (Wolraich, Siperstein & Reed, 1991) when an infant with Down Syndrome, requiring surgery for oesophageal atresia, was allowed to die by the attending doctors in accordance with the wishes of the parents. Children with Down Syndrome were often subjected to institutionalisation where they were forcefully sterilised or used for experimental trials, such as vaccine testing without consent (Michaels, 1990).

In the last two decades, there has been a trend toward treatment of early correctable congenital heart defects, de-institutionalisation, retention of children in the home environment and strong advocacy for free special education for all children with disabilities. The emergence of this current philosophy of management, together with the advocacy for the adoption of a "mainstream" education system for these children has placed great pressure and responsibility on the medical profession to be familiar with the developmental and mental capabilities of these children (Wolraich et al, 1991). The physician, armed with the knowledge of long term problems of these children is well placed to guide the parents in their delivery of home care and school

programmes (Melyn & White, 1973).

However, whilst there has been an improvement in their life span, problems attendant to children with Down Syndrome spending more time in parental care are beginning to emerge. First, the family unit itself has become vulnerable (Gath, 1992; Johnston, 1991). Children with Down Syndrome develop behavioural and medical problems. Second, the effects on the marital relationship of the parents are often serious, as they experience more stress when they try to cope with problems relating to nurturing a disabled child. Further, other siblings are also affected. Behavioural problems rise to the surface because of sibling rivalry with the affected child for parental love and attention. It has therefore become necessary for service delivery in Down Syndrome not to be directed only to the individual child, but, that consideration be given to the child's eco-systems, such as the family and the community (Katz, 1995).

Therefore the broad management of Down Syndrome includes the areas of prevention, early detection with counselling, management of physical ailments, psychiatric illnesses and adolescent handicaps. If doctors are to be part of the network for the assessment and of the support services in the community, they should be equipped to dispel the negative images of this syndrome (Gath, 1990; Johnston, 1991; Grieve & Mphelo, 1993).

1.1.2 THE PURPOSE OF THIS STUDY

The purpose of this study was to investigate the benefits of a home programme as part of the management of infants and children with Down Syndrome, between the ages of six months and six years in the Witwatesrand area. The home programme used was S.T.A.R.T. - Strive Towards Achieving Results Together. The developmental level of these children was assessed by measuring their general or developmental quotient (D.Q.) on the Griffiths Development Scales (Griffiths, 1970; 1984). The results of the treatment group was then compared with the results of a similar group of infants and children who had not participated in the S.T.A.R.T. programme. Furthermore it was assessed whether using the S.T.A.R.T. programme was associated with any benefits for the parents or caregivers.

1.2 THE DIAGNOSIS AND PROBLEMS ASSOCIATED WITH DOWN SYNDROME

1.2.1 DIAGNOSIS OF DOWN SYNDROME

Diagnosis of Down Syndrome can be made in the following ways:

1.2.1.1 Clinical Diagnosis

Down Syndrome may be diagnosed at birth. On physical examination the neonate shows the following clinical features (Rudolph & Hoffman, 1982; Richards & Reed, 1991; Christianson, 1996; Kwong & Wong, 1996)

1.2.1.1.1 **Head and neck**. The infant has a small head, flattened occiput and a recessive nose. There is an upward slant of the eyes with epicanthal folds. The eyes show Brushfield spots. Small low set ears is another common feature. The tongue is large and protruding. There is a broad stocky neck with a low set hairline and loose skin folds.

1.2.1.1.2 **Chest**. A pigeon chest, with an increased anterior-posterior diameter is common.

1.2.1.1.3 **Skin.** The skin is mottled and the hair may be thin and fair.

1.2.1.1.4 **Digits**. The digits of the hand and foot are short, giving a stubby appearance. The fifth digit shows some incurving. A single palmar crease is often found.

1.2.1.1.5 **Neurological**. There is general hypotonia.

A confirmation of the karyotype is always mandatory (Thapar, et al, 1994; Vyas, 1994).

1.2.1.2 **Pre-Natal Diagnosis of Down Syndrome**

Viljoen (1996) describes pre-natal diagnosis as a process by which congenital or hereditary abnormalities are detected in the unborn child. Prenatal diagnosis should be offered to all pregnant women. It is definitely indicated in 7 - 8% of all pregnancies in South Africa (Kromberg, 1989). Prenatal diagnostic procedures are divided into two

categories (Viljoen, 1996) namely:

1.2.1.2.1 **Prenatal Screening Tests**

These refer to measurement of biochemical markers in the venous samples of blood taken in the mid trimester, in order to detect foetal congenital abnormalities. These tests are non-specific and may only mark pregnancies at a potential higher risk.

1.2.1.2.1.1 **Triple Test.**

Antenatal screening of maternal serum for Down Syndrome is effective in practice and can be readily integrated into routine antenatal care. It is cost effective and it is a better predictor than selection for aminiocentesis on the basis of maternal age only (Wald, Kennard, Densem & Butler, 1992). Younger women who are positive may opt for an amniocentesis, while older women who are negative are spared the risks associated with the procedure. The test involves calculation of foetal risk, based on four parameters as shown below: (Table III).

TABLE III. TRIPLE TEST COMPONENTS AND THEIR CONTRIBUTION

Component	Detection of Down Syndrome
Maternal age only	30%
Maternal age + AFP	40%
Maternal age + AFP + HCG	60%
Maternal age + AFP + HCG + E	65 – 70%

(AFP - a - fetoprotein, HCG - Human chorionic gonadotrophin, E - eostriol)

A low AFP, an elevated HCG and a low oestriol are associated with increased risk of foetal Down Syndrome, and identified more than 61% of positive results in a study described by Viljoen (1996). These parameters are population specific and a normal range must be determined for any community. If the risk of foetal chromosome abnormalities exceeds the risk of amniocentesis, then amniocentesis should be offered.

1.2.1.2.1.2 **Foetal Sonography**

An ultrasound finding suggestive of the following should be an indication for an amniocentesis: (Viljoen, 1996).

(i) Short femur and humerus.

(ii) Thick nuchal fold.

(iii) Polyhydraminios.

(iv) Duodenal atresia.

(v) Cardiac abnormalities.

1.2.1.2.2 **Prenatal Diagnostic Tests**

1.2.1.2.2.1 **Amniocentesis**

Although this is a relatively safe procedure, it is associated with a risk factor of about 0.5%. It is not usually offered to women below the age of thirty five, unless other risk factors exist, or after 16 weeks gestation.

Indications for amniocentesis in the diagnosis of Down Syndrome are:

(i) Previous child born with Trisomy

(ii) Balanced translocation carrier state.

(iii) Positive maternal screening (Triple Test)

(iv) Suggestive abnormal foetal ultrasound.

(v) Older women at risk

1.2.1.2.2.2 **Villus Sampling**

Villus sampling, or chorionic villus biopsy, generally performed between the 9th and 12th weeks of gestation results in an earlier availability of results than is the case with amniocentesis. There are, however, well described problems attendant with this procedure, such as

(i) Villus sampling is technically more difficult

(ii) Villus sampling is associated with a higher miscarriage rate - that is, 2-5% as opposed to 0,5% for amniocentesis (Vijoen, 1996).

1.2.2 PROBLEMS OR COMPLICATIONS ASSOCIATED WITH DOWN SYNDROME

1.2.2.1 MEDICAL PROBLEMS IN DOWN SYNDROME

1.2.2.1.1 Problems in the Neonate

The most common problems in this age group are:

1.2.2.1.1.1 **Congenital Heart Disease**

40-50% of children with Down Syndrome are born with congenital heart disease, (Christianson, 1996) the most common of which is endocardial cushion defect (Hayes & Batshaw, 1992; Kwong, 1996). Others include atrial septal defect, patent ductus arteriosis and ventricular septal defects. The infant mortality of children with Down Syndrome is intimately related to the presence of specific types of heart defects. The use of surgery in these conditions is a great factor in the reduction of infant mortality in Down Syndrome.

1.2.2.1.1.2 **Gastro-Intestinal Problems**

Most problems of the gastro-intestinal tract are related to obstruction and will present in the early neonate period. The most common abnormalities include tracheo-oesophageal fistula, pyloric stenosis, imperforate anus and duodenal atresia, all of which are surgically correctable (Carey, 1993; Kwong, 1996)

1.2.2.1.1.3 **Other Problems**

(a) **Hypothyroidism**. There is an increased incidence of congenital hypothyroidism in the newborn with Down Syndrome (Hayes & Batshaw, 1993). The precise mechanism of this condition is not understood. Newborn screening is strongly recommended.

(b) **Leukaemoid Reaction**. This may develop into full blown leukaemia, but generally it resolves spontaneously in the first month of life.

(c) **Dysplastic Hip**. Congenital dislocation of the hip are one of the commonest causes of difficulty in mobility in the Down Syndrome children. It is said to be associated with joint laxity and hypotonia in these children.

1.2.2.1.2 **Childhood Medical Problems in Down Syndrome**

1.2.2.1.2.1 **Physical appearance**. The facial features of Down Syndrome are typical and are present in all cases albeit in varying degrees of severity. Many of the features are said to occur as a manifestation of "incomplete" rather than "abnormal" embryogenesis (Hayes, 1992). Facial plastic surgery has been performed with varying success and not without ethical concerns. (Cunningham, Turner, Sloper & Knussen, 1991)

1.2.2.1.2.2 **Hearing loss**. Conductive hearing loss in seen in 40% - 60% of patients with Down Syndrome, at least half of whom have bilateral impaired hearing and is assumed to be due to chronic serous otitis media. Mid-facial hypoplasia is found to be a contributory factor in the aetiology of otitis media. Neuro-sensory loss becomes evident later and affects 20 - 30% of patients with Down Syndrome. Routine audiology evaluation before eight months of age and yearly follow up examinations are recommended.

1.2.2.1.2.3 **Thyroid disorders**. 13 - 45% of older individuals have biochemical evidence of hypothyroidism.

1.2.2.1.2.4 **Ophthalmic problems**. The commonest eye problems in children with Down Syndrome are refractive errors, primarily myopia. In addition there is a high prevalence of nystagmus (35%), strabismus (50%), cataracts (13%), blepharitis and nasal lacrimal duct obstruction and Brushfield spots.

1.2.2.1.2.5 **Dental issues**. A very common occurrence in children with Down Syndrome is delayed tooth eruption, the first tooth only erupts after 20 months. Malocclusion, gingivitis and periodontal disease is associated with dental carries.

1.2.2.1.2.6 **Oro-Pharyngeal Problems**. Sleep apnoea and snoring occur as a result of an under developed nasopharynx with tonsillar

and adenoidal hypertrophy. Prolonged sleep apnoea results in pulmonary artery hypertension and cor pulmonale.

1.2.2.1.2.7 **Musculo-Skeletal Abnormalities**. Hypoplasia and joint laxity are found commonly to result in the delay of development of gross motor skills, such as sitting (average age one year), walking (average age two years),

1.2.2.1.2.8 **Joint Dislocation**. Dislocation at the hip and knee are common occurrences in children with Down Syndrome. Bones of the upper spine and neck are under-developed, resulting in atlanto-axial subluxation in 10 per cent of cases, spinal cord compression at 0.1 per cent. Other symptoms are clumsiness, head tilt, limping, refusal to walk and limb weakness.

1.2.2.1.2.9 **Haematological Abnormalities**. The risk of the development of acute lymphoblastic or non-lymphoblastic leukaemia in the children is 1%.

1.2.2.1.3 **Medical Problems in the Adolescent with Down Syndrome**

Mortality in early and mid adulthood in the population of Down Syndrome is relatively low (Fryers, 1986). Good general health is reported in 80 per cent or more of young adults with Down Syndrome (Holmes, 1988).

1.2.2.1.3.1 **Skin problems** are frequent in this age group and affect about one quarter of individuals, and includes thinning sensitive skin and rashes and alopecia.

1.2.2.1.3.2 **Epilepsy** occurs as generalized tonic-clonic seizures in the third and fourth decade in people with Down Syndrome (Smith & Wallace, 1982; Stafstrom, Patty, Gilmore & Wisniewski, 1991).

1.2.2.1.3.3 **Congenital heart disease** is found in 15 - 20% of the adolescents with Down Syndrome.

1.2.2.1.3.4 **Cancer**. The incidence of cancer in this age group appears to be similar to that in the general population, apart from an increased risk of childhood leukaemia.

1.2.2.1.3.5 **Obesity** is seen commonly in the adolescent child with Down Syndrome, although there is no known actual documentation on this subject.

1.2.2.1.3.6 **Visual and Hearing** problems. The incidence of these problems varies considerable but poor vision requiring spectacles is common and hearing problems were estimated at 10 - 20% (Carr, 1993).

1.2.2.1.3.7 **Sexuality.** Studies of sexual development in both male and female persons with Down Syndrome have shown no major differences in the sequential emergence of primary and secondary sex characteristics and increase in sex hormones from the norm (Cunningham, 1984). Sexual drive, although somewhat delayed, is not significantly impaired in these

individuals.

1.2.2.2 DEVELOPMENTAL PROBLEMS

Infants and children with Down Syndrome have a significant developmental disability in the areas of physical growth, gross and fine motor development, cognitive and mental development, speech and language (Cunningham, 1984).

1.2.2.2.1 **Growth Deficiency in Down Syndrome.**

Cross sectional studies in growth development in children with Down Syndrome have consistently shown growth deficiency, beginning pre-natally and continuing until the children are between three and five years of age (Cronk, 1977). Average length and weight are reduced two Standard Deviation (SD) from the mean by the age of five years. In a Boston study by Cronk (1977) involving ninety infants with Down Syndrome, data for growth charts were obtained that are useful in the assessment of children with Down Syndrome (see Figure 2). The results in the graph show that the growth patterSyndrome (see Figure 2). The results in the graph show that the growth normally developing children. In this study (Cronk, 1978) the mean measured recumbent length at birth in children with Down Syndrome was reduced by 0.5.SD from the mean of the control group and at three years by 2 SD below the control mean. The growth velocity is also deficient and graphs fall below the expected range and even below the third percentile. The mean curve for

weight reflects a general tendency to be overweight in these children. Although the ini the initial headcircumference in neonates with Down Syndrome have been shown to be often within normal limits, it does decline and cross percentiles over the first few years. (Wisniewski, 1987; Kwong, 1996). In a subsequent study, Wisniewski (1990) reports that the brain weight of infants with Down Syndrome showed a 20-50% reduction by mid-infancy.

Standardised circumference growth charts for assessing development are presently available for use (Gath, 1992).

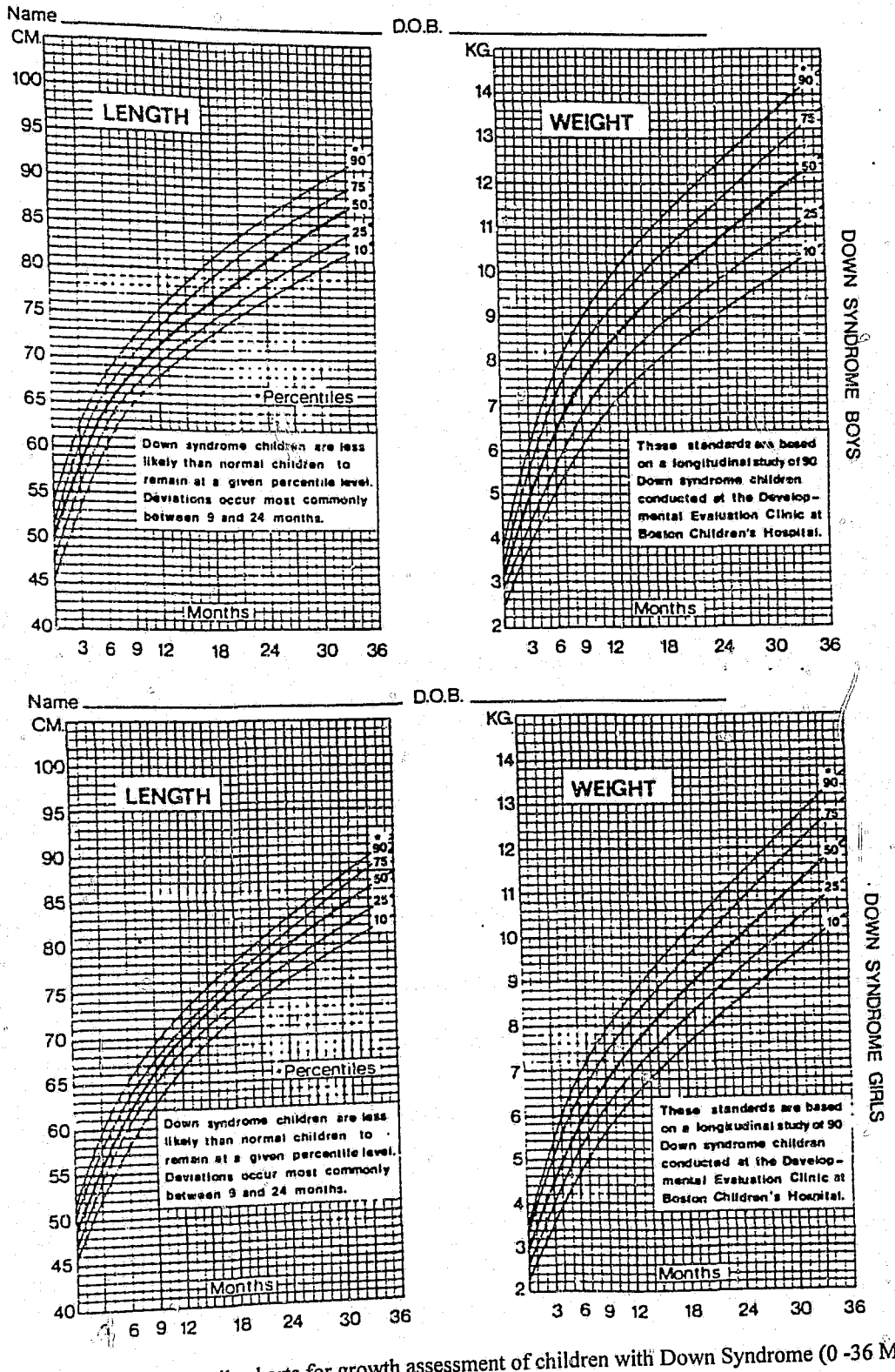


Figure 2: Percentile charts for growth assessment of children with Down Syndrome (0 -36 Months)

1.2.2.2.2 Development of cognitive or intellectual function.

Wisniewski (1990) reports that Down Syndrome individuals show developmental delay in language and intellectual functioning secondary to central nervous system abnormality (cortical dysgenesis). In the same study it was also found that the occipito frontal circumference (OFC) and brain weight (BW) of newborns with Down Syndrome are often at 2SD below normal, but that differences increase with age, especially after 3-6 months. The OFC and BW are often at -2SD below the norms (Wisniewski, 1990).

Although studies have demonstrated a general decline in the Intelligence Quotient(IQ) in children with Down Syndrome in infancy to late childhood (Melyn & White,1976; Carr, 1993; Neser, Molteno & Knight, 1987; Kwong, 1996), it has also been shown that children cared for at home were generally more advanced than those cared for in an institution, possibly, because institutions are unable to provide the same level and quality of individual attention (Centerwall & Centerwall, 1958). Further, it was found that the most significant period for effective home care was the first three to five years of life and that the advantage would be maintained for many years (Cunningham, 1986). Rapid development was also reported in the children with Down Syndrome who were given early systematic stimulation and that positive long term effects were elicited, and in particular, development of academic skills, such as reading and arithmetic for children with Down Syndrome

(Cunningham & Morgan, 1984; Cunningham, 1986; Thapar, 1994; Carr, 1993).

In their assesment of 55 pre-school children (mean 38month) with Down Syndrome living in Cape Town, using the Griffiths scale of Mental Development, Nesser, Molteno and Knight (1989) also showed a significant decrease in Developmental Quotient with increasing age, especially of speech and language abilities. They further more found no significant relation of the child's developmental abilities, and maternal age, sex social class or race, but children with one or no siblings functioned better than those with two or more. The latter was thought to reflect the time parents had to interact with their handicapped child. Children in structured care-facilities tend to perform at a higher level than those cared for solely at home.

The trend of global IQ or rate of development of children with Down Syndrome from 0-36 months is shown in Figure 3, and it can be seen that the most rapid decline in the rate of development occurs within the first three years (see Appendix C - Development Milestones in Down Syndrome). The rate of development drops off around the first six months of the developmental course. There seems to be a significant deterioration in the sensory motor co-ordination, such as eye-hand co-ordination, co-ordination of balance and muscle control, as is evidenced by the difficulties children with Down

Syndrome experience in picking up objects and learning to sit up. Studies confirming poor integration of sensory information such as directional judgement, propioceptive and visual reference have shown that activities such as drawing and copying were deficient in children with Down Syndrome (Connolly, Morgan, Russell & Fulliton, 1993).

Postural control problems were identified by Shumway-Cook and Wollacott (1995) who showed that postural responses were slow in children with Down Syndrome between the ages of one to six years. Several workers stressed the importance of the relationship between the evolution of postural reactions and subsequent acquisitions of milestones in infants with Down Syndrome (Rast & Harris, 1985). Carr (1993) suggested that all motor assessment of children with Down Syndrome should include an evaluation of postural reactions and movement and that intervention programmes should promote the development of postural reactions, in order to enhance the onset of developmental milestones.

The dips in the rate of development in Figure 3 reflect specific difficulties for many children with Down Syndrome, making their development appear even slower. The first downward swing or dip, is followed by an upswing which represents a rapid increase in development. This heralds the next stage of playing and exploring objects by sucking, banging things, holding of objects,

passing objects from hand to hand. It depends on the ability of the baby to sit up fairly well, reach out and grasp and is usually achieved at the age of nine months. Between the age of ten to thirteen months another stage of slow rate of development ensues, when the children have to learn that objects are different, have special qualities and that the children themselves have to process this information during play. They need to practice and consolidate new skills. It would seem that this period of consolidation takes longer in children with Down Syndrome. Rast et al (1994), in their study of mastery of motivation in children with Down Syndrome, found that these children had a lower level of task engagement and displayed less causality pleasure than normally developing children.

At the beginning of the second year, most normal children are able to say at least three to four single words, solve simple problems like putting strings together, display goal directed behaviour in playing, imitate single actions and noises. All these need memory and a definite attention span. This developmental stage appears to be delayed in children with Down Syndrome (Cunningham, 1986), and they reach a significant plateau between 18 and 24 months of age. Therefore, information would have to be presented at a slower rate and in small amounts when rehabilitating children with Down Syndrome.

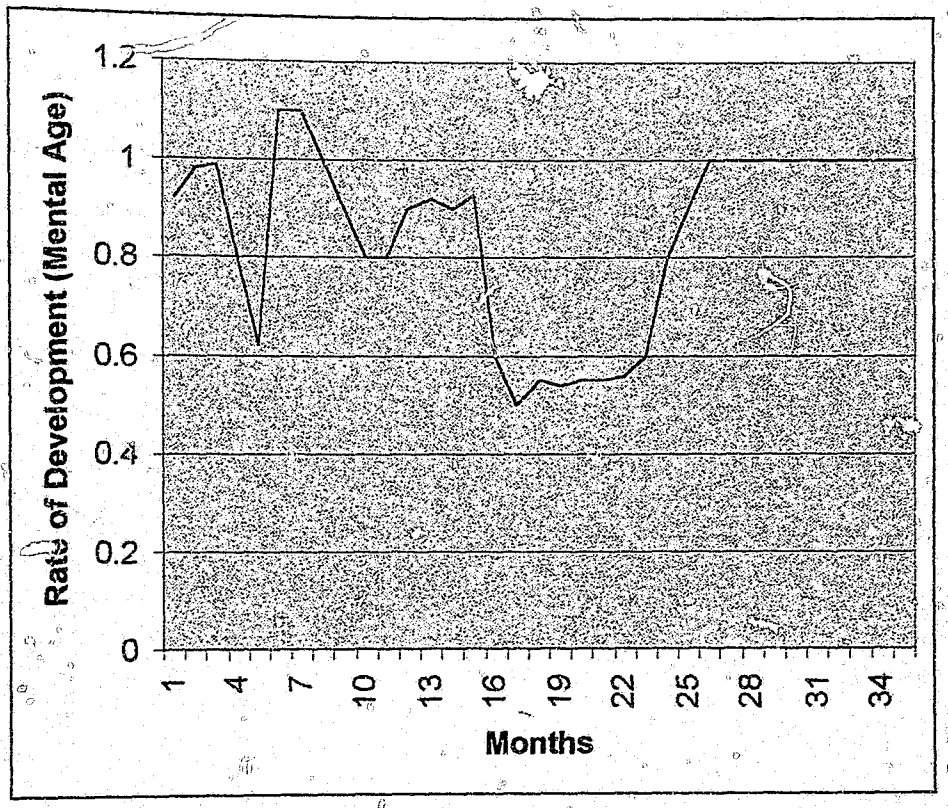


Figure 3: Changes in mean developmental rates: mental age

(Source: Cunningham, 1986: 31)

1.2.2.3 Speech and language development

Studies in speech and language development frequently report severe productive language delays in children with Down Syndrome (Collacott, 1993; Drash, Raver & Tudor, 1989; Hayes & Batshaw, 1993). These delays appear to persist across the life span and restrict communication competence. Babbling was investigated by use of the Illinois Test of Psycholinguistic Abilities (ITPA) and was found to be similar to that of normally developing children, but at a lesser frequency (Dodd, 1972; Steffens, Oller, Lynch &

Urbano, 1992).

Linguistic imitation by normal children of their mother's speech, was said to facilitate acquisition of new vocabulary (Clark, 1974, 1977). Sokolov (1992) showed that children with Down Syndrome displayed imitation and turn taking of their mother's speech to a lesser degree than normally developing children, but that the nature of the difference was more noticeable when the mean length utterance (MLU) was low. Bilonsky and Share (1965) found on ITPA tests that children with Down Syndrome performed better when visual perception and manual expression were utilised, than when auditory and visual skills were required. Research has shown weaknesses with communication abilities in children with Down Syndrome involving specifically the expressive ability (Dykens, Hodapp & Evans, 1994; Miller, 1988).

An examination of articulation problems in children with Down Syndrome by Zisk and Beiler (1967) has highlighted some potential predisposing factors, such as an underdeveloped upper jaw, which results in malocclusion and interferes with the production of explosive sounds like "s" "f" "t". The other factor is the apparent tongue hypertrophy as a result of a small cavity. Poor oral-motor function which is possibly due to hypotonia, results in poor sucking and chewing. The presence of hearing loss is also regarded as a contributory factor in articulation problems. Profound hearing impairment was found to

produce delays in canonical babbling (Oller & Eilers, 1988).

1.2.2.4 Development of Adaptive Behaviour in Down Syndrome

Adaptive behaviour is defined by the American Association of Mental Deficiency as the "quality of everyday performance in coping with everyday demands" (Cunningham, 1988). The profiles of adaptive function are identified by Dykens et al (1994) as first communication, which consists of sub domains of expressive, receptive and written language skills. Second, daily living skills which deal with behaviour related to personal grooming and domestic chores and functioning in the community. Third, socialization, which is associated with behaviour involved in getting along with others, playing and coping with environmental demands. It was pointed out in earlier studies that children with Down Syndrome had weaknesses in expressive communication abilities related to functions in the other areas and that this deficit became pronounced as children developed in mental age (MA). The developmental course of adaptive functioning in children with Down Syndrome was found to be stalled, and the children showed difficulties in maintaining certain levels of performance within particular types of tasks (Hodapp, 1990). In addition such slowing in development was found when infants with Down Syndrome traverse specific stages of the Piagetian sensori-motor development (Cichetti &

Beighl, 1990), exhibiting an advance-plateau pattern of adaptive development during the early and mid childhood years. Therefore, a more aetiology based intervention programme and more precise training programmes in adaptive skills needs to be implemented in the rehabilitation of the children with Down Syndrome.

1.3 THE TREATMENT PROGRAMME (S.T.A.R.T.)

1.3.1 Early Intervention Programmes

Early intervention is a multi disciplinary service provided for developmentally vulnerable and disabled children from birth to three years (Russel, Kirby & Swanson, 1993; Ramey, Bryant & Wasik, 1992; Simeosson, Cooper & Scheiner, 1982; Collin, 1995; Botha, 1987). Such service is also concurrently made available to affected families. As Meisels and Shonkoff (1990: vii, as quoted by Kara, Harilaou, Masoga, Phiri & Barrett, 1995: 16) state, early intervention:

> "is designed to enhance child development, minimize potential delays, treat existing problems, prevent further deterioration, limit acquisition of additional handicapping conditions and promote adoptive family functioning. The goals of an early intervention are accomplished by providing developmental and therapeutic services for children and support of their families".

There are several factors which might determine the effectiveness of an early intervention programme, such as the type of the intervention, the intensity of a child's disability and the age at initiation of the programme. It has been found that enrolment of children with Down Syndrome before the age of 6 months is often associated with significantly better outcomes than enrolment at a later age (Shonkoff & Hauser-Cramp, 1987).

According to the World Health Organisation (1989) the rate of disability in developing countries is as high as 7 to 10%. For South Africa, the implications of this ratio are that, given the 1991 census figures, out of 66097 children in Soweto between the age of 0-4 years 6609 children could be disabled. This large number of children with special needs, coupled with the lack of resources and the shortage of professionals in the medical and allied professions, has compounded the problem of services for early intervention (Katz, et al, 1995).

The paucity of services for pre-school mentally disabled children in South Africa, especially among African and socio-economically disavantaged children was highlighted by Cartwright, as quoted by Katz et al (1995), where they found that the availability of services in the Witwatersrand area was totally unbalanced, as reflected by the following percentages of services provided for each population group: white - 69.7%, Indian - 26%, Coloured - 9.3% and

African - 7.6%. In rural and poor areas, very few individuals benefitted from services which were provided. The World Health Organisation recommends that the provision of services to increased numbers of people could be achieved through community-based rehabilitation where the responsibility of the rehabilitation is taken by the individual, the family and the community. (Collins, 1995; Botha, 1987)

There are two major benefits of early intervention programmes, those experienced by the parent and those aimed at the child with special needs. As far as the parents are concerned, the negative effects of having a child with disability are well described in literature. Such a parent may have feeling of isolation, depression and helplessness (Solarsh, 1986). Participation in programmes helped to reduce feelings of helplessness and empowered the parent to work effectively with the child. The S.T.A.R.T. programme is an early intervention programme intended to support both parents and their children with special needs. Katz et al (1995) studied the perceived effects on parents who had regular contact with the S.T.A.R.T. programme and concluded that the most helpful aspects included support, positive reinforcement and motivation to continue. Katz et al also found that the coping capacity of mothers improved in 100% of those who were involved with the S.T.A.R.T. programme - and 70% of the mothers felt their expectations were met.

Previous studies indicated that children with Down Syndrome received very little specific training and attention in their early life (Carr, 1993). Yet, they appear to have a potential for learning, which could be developed by using appropriate methods (Cunningham, 1988). Reynders and Horrobin (1990) concluded that children with Down Syndrome were not only trainable in terms of their function, but also educable. It would appear though, that the importance of early intervention lies in the prevention of the decline of intellectual and adaptive functioning of these children (as described by Carr, 1970, in untreated infants with Down Syndrome). This benefit was documented whether a child-focused Early Intervention Programme (EIP) (Connolly, Morgan & Russel, 1984), was used or a maternal training programme (Bidder, Hewit & Gray, 1975). Similar gains in early intervention for children with Down Syndrome were found by Hanson (1979) after two years of structured intervention programme. Piper and Pless (1980) could not demonstrate any positive effects of their early intervention programme, although these conclusions were criticised by Bricker, Carlson and Schwartz (1981) because of lack of standardisation of the intervention, poor definition of objectives and short periods of intervention.

The importance of early intervention programmes (EIPs) has been demonstrated by a number of workers in various countries. Some of these

programmes are used as community-based early intervention programmes, the general purpose of which is as defined above by Meisels and Shonkoff (1990). Examples of such programmes include Step-by-Step in Guyana (O'Toole, 1989), utilising volunteers drawn from a wide range of occupations; the 3D Programme (Jamaica); and Project Projimo in Mexico (Katz et al, 1995). It is quite evident, therefore, that the concept of early intervention is widely held as an important way to deal with the developmental problems of children. A home programme, like S.T.A.R.T., which optimizes domestic care in children with Down Syndrome should be a valuable tool in the reduction of the need for institutionalisation of such children.

1.3.2 Major Characteristics of the S.T.A.R.T. Programme

The S.T.A.R.T. programme is a "home grown" programme which emerged in response to the request of families who had to cope with handicapped children. The S.T.A.R.T. (Strive Towards Achieving Results Together) programme is a relatively new programme, developed in 1981, and is especially directed at those handicapped individuals with special needs, without access to a suitable early intervention service. It was compiled through a combined effort of health professionals including speech therapists, psychologists, paediatricians and social workers mainly from the University of the Witwatersrand. As such, the S.T.A.R.T. programme is equipped to serve children with a variety of disabilities, using a structured approach which is adaptable and can be used in

urban as well as rural settings. It was primarily designed for use by parents, but may also be used by professionals in isolated areas. The S.T.A.R.T. initiative, together with Toy Library, is part of community outreach service of the Sunshine Centre Association - an association whose aim is to provide a range of early intervention services for the mentally handicapped children and their families. It therefore compliments the philosophy of primary and community based health care of the present Government. The S.T.A.R.T. programme encapsulates two vital principles:

(a) It is essentially a parent training programme. The most important reasons why early intervention programmes should involve parents are described by Eldelstein (1981):

(i) The number of handicapped children requiring specialised attention far out numbers the professional resources available, particularly in the pre-school years;

(ii) Because of their close and embracing relationship with their children, parents can play a major role in their children's development, particularly in the formative years. Therefore, parents need to have a sound knowledge about the sequence of normal development for the successful rehabilitation of their handicapped children; and

(iii) Parents are natural teachers of their children, as they are always

with them in their own environment, in the home and in the community.

(b) The S.T.A.R.T. programme is an integrated programme which takes a holistic view of the child from 0 - 18 months, which stresses the partnership between the programme, the parent, caregiver and the community. It involves information relative to child development, family reactions to developmentally delayed children and how to assist the parent or caregiver in the direction of the use of the material.

The programme consists of a comprehensive package of material, including guidebooks and check lists for evaluation of children in all developmental areas such as gross motor skills, language skills, fine motor development and activities of daily living. In addition, appendices on the making of toys and equipment, stimulation of the hearing impaired and ways of handling the severely handicapped child are all included in the package.

1.4 THE GRIFFITHS DEVELOPMENT SCALES (GRIFFITHS, 1970; 1986)

1.4.1 Background and Major Aspects of the Griffiths Development Scales

The importance of early assessment of potentially handicapped children cannot be over-emphasized. Holt (1979: p151) gave a very succinct summary of this

view when he stated:

> "Any child who is suspected of having a congenital defect or deformity, a deviation of development, a medical disorder which may produce continuing disability, an impediment to educational progress or social activities or any deficiency of opportunities is a potentially handicapped child and should be assessed."

The Griffiths Development Scales were developed in Great Britain for use with children from ages 0 to 8 years, and so provides norms for infants and preschool children. Ruth Griffiths, in both her books "The abilities of Babies" (1986) and "The Abilities of Young Children" (1986), gives a report based on the research which she did on 2000 children. She developed the scales by observing the behaviour of children in their natural habitat. The test comprises global and sub-scale developmental quotients (DQs) so that comparisons can be made of specific abilities. The sub-scale DQs are particularly useful in the assessment of children with Down Syndrome since their scores extend to below 50 (Neser et al, 1987).

The Griffiths scales are considered "culture friendly" and have been used outside Great Britain for research projects in countries like Columbia, Lebanon, France, Germany, Canada and South Africa (Allan, Luiz, Foxcroft, Unpublished data). The Griffiths Scales have also been successfully used

internationally in longterm developmental studies (Bidder, Bryant & Grayet 1975; Hindley, 1965; Eaves, 1970; Elliman, 1985; Piper & Pless, 1980; Smith & Wallace, 1982; Holt, 1974, 1979).

Having been introduced in South Africa in 1977, Griffiths Scales have been used in various studies related to the assessment of developing young children (Goodman & Rothberg, 1985; Helmes, 1983, Neser et al, 1989)

1.4.2 Applicability of the Griffiths Development Scales

In a study by Evans (1992) it was shown that in the components of language, fine motor and activity for daily living (ADL), the Griffiths scales were congruent with those of the S.T.A.R.T programme. The Griffiths scales are well researched and have been found by Helmes (1983) to be culture fair and correlate highly with the Junior South African Individual Scale (J.S.A.I.S.).

Bowen et al (1996), embarked on a study to assess the relationship between Griffiths Development Scales at 1 year and 3 years, and Stanford-Binnet intelligence scale. It was found that the 3 year Griffiths Scale developmental quotient (DQ) is a good predictor of 5 year Stanford-Binnet intelligence(IQ) in extremely low birth weight (ELBW) children, and can be used to identify children who may benefit from intervention programs prior to school entry.

CHAPTER 2: METHODOLOGY

2.0 INTRODUCTION

In this chapter the methodologies such as research design, the target population, study sample and size, procedures, instrumentation and data analyses of this study are presented.

2.1 RESEARCH DESIGN

This is a multi-method (parental questionnaire, medical and neurological examinations and developmental assessments) cross sectional, ex post facto, (planned and performed after the treatment had been instituted) study, comparing two groups of children with Down Syndrome, - treatment and control, matched on the predetermined criteria of age, sex and socio-economic status, on the Griffiths Development Scales (Griffiths, 1970, 1986). Although the control group was not matched, it was anticipated that they would be similar to the treatment group for criteria such as age, sex and socio-economic status.

2.2 STUDY POPULATION

The study population comprised all children with Down Syndrome in the Republic of South Africa, whilst the accessible population for this study were all infants and children with Down Syndrome who resided in the

Witwatersrand area of the Republic of South Africa at the time of this study and in particular Soweto, between the ages of 6 months and 6 years. However, for logistic and practical reasons, it was not possible for the researcher to establish the actual number of children with Down Syndrome residing in the Witwatersrand area, since there were too many authorities and centres (i.e. pre-schools, health centres, clinics, hospitals, etc.) having their own records concerning Down Syndrome. But it can be presumed, based on an incidence figure of plus/minus 1.3 per 1000 live births (see Christianson, 1996) that there could be about 86 children of all ages with Down Syndrome in the community. However, the researcher feels that the research design used in this study was credible enough to help her to address the stated research questions.

2.3 STUDY SAMPLE

The study sample was recruited from a list of Down Syndrome patients enrolled in the Speech Therapy Department at Baragwanath Hospital, Soweto which serves a large number of Down Syndrome children from urban, rural and peri-urban areas.

To enter the study all children had already been diagnosed to have Down Syndrome clinically and biochemically and had been accepted by the Speech Therapy Department for the Down Syndrome treatment group.

2.3.1 The Treatment Group

The criteria which were used to qualify subjects for participation as members of the treatment condition are summarised in Table V below:

TABLE V: SAMPLE ELIGIBILITY CRITERIA FOR EXPERIMENTAL GROUP

1.	Confirmation of diagnosis
	(i) Clinically
	(ii) Biochemically
2.	Age: Between 6 months and 6 years
3.	Participation in the START programme for at least 6 months
4.	No other intervention programme

Initially, thirty subjects (n=30) who had participated in the S.T.A.R.T. programme were considered for possible inclusion in the study; but of these, 7 patients could not be traced, two patients were admitted in the paediatric medical wards and so could not be tested and one died in the wards from complications of congenital heart disease and so was lost to the study. In the final analyses, therefore, the treatment group comprised of 20 subjects.

2.3.2 The Control/Comparison Group

A modified control or comparison group of twenty children known to have Down Syndrome was selected from the waiting list of the Department of

Speech Therapy or from families who had elected not to take part in the intervention programme. These children had to fulfil the same inclusion criteria as the treatment group, except that they were not to have received any intervention similar to the treatment group.

Because all patients were recruited from the same socio-economic group and geographical location, it was assumed that the distribution of economic status would be similar in both groups. Yet some families who elected not to participate in the intervention programme differed in some important characteristics from those who did, such as the level of impairment of the child, family organization, paternal or maternal educational status. These factors were specifically noted in the accumulation of data. The research period was five months extending from February to June, 1994.

2.4 INSTRUMENTATION

2.4.1 Treatment of Condition

During the time of this study children with Down Syndrome at Baragwanath Hospital were, from birth to three years of age, referred to the Speech Therapy Department from physicians, social agents and parents, for possible enrolment to the S.T.A.R.T. programme. Eligibility for the programme was based on the parent/caregiver being able to bring the child in and follow through with activities at home. Early referral is a basic requirement for early intervention

programmes. The programme was structured in such a way that the parents were the primary programmers for their children since the programme had to be carried out at home for at least two hours per day - one hour in the morning and another in the evening. The programme also sought to promote family stability through group experience for the parents and/or caregivers.

The intervention programme, S.T.A.R.T., consisted of weekly centre-based therapy sessions of half a day per week, until the age of three years. The child's condition was evaluated and the child then placed on an individualised home programme. Use was then made of the materials provided, which comprised checklists, integrated programmes, activity sheets and appendices to guide and teach the parents. The child's level of development was plotted on appropriate checklists. The checklists then directed the user to recommended activities that could be carried out on the child.

Parents and children were seen in group-sessions with other parents. The sessions were informative to the parents. The information given was on motor, sensory, play social activity, language development and self help skills. The activities were individualised to each child according to developmental profiles. Normal sequential development of the child was explained, as many parents were unaware of what to expect of their children. Physical therapists encouraged gross motor activities, like muscle strengthening, and gross motor

stimulation. Nurses directed activities of sensory stimulation, including simple games like pick-a-boo. Parents were encouraged to discuss their problems and share ideas with other parents, and also to belong to relevant associations, such as the Sunshine Association for Parents of Children with Down Syndrome. The home programme was conducted during daily routines, for example, bath time and nappy time; or specific activities would be incorporated into daily routines, like mother supervision during meal times. Re-evaluation of the progress was done regularly to give a realistic picture to the parents or teacher/counsellor, then a new and appropriate programme would then be formulated. Various teaching methods, like modelling, shaping and re-enforcement were used. Overall, emphasis was placed on the continuous day-to-day stimulation of the child and not limited to the sessions given in the centre.

2.4.2 TESTING PROCEDURES

Letters (See Appendix E) were sent to all parent-child pairs to inform and invite them to participate in the study. Five patients were scheduled per day. All parents (mothers and fathers where applicable) and caregivers were informed of the nature and objective of the study, whilst being assured of the confidentiality of the results by making use of a coding system (see Appendix G). Written informed consent (see Appendix F) was obtained from the parents of the children who participated in the study. The testing time per participant

was two to three hours. Tests and interviews were administered in the vernacular by a trained researcher in a quiet room in all cases.

2.4.2.1 Administration Procedure for the Griffith's Development Scales (Griffiths,1970;1986)

The child's development status was determined using the Griffiths' Development Scales (See Appendix D). This was performed before the interview so that the researcher was blind as to whether or not the child had attended the S.T.A.R.T. programme. The Griffiths' Development Scales were administered and scored according to the standardisation procedures laid-down by Griffiths (1984). A few items had to be adopted for local use, for example, "cent" for "penny", etc. Instructions were given in the language acceptable or easily understood by the child. (Reliability for test scores had been established through studies of inter-rate reliability and test – retest reliability at 90% and 86% respectively). If a child became ill or tired before completion of the test, the test was rescheduled for another time. The interviews and testing were carried out by the researcher (I.N.S) who has had appropriate training in the administration of the Griffiths' Developmental Scales.

2.4.2.2 Interviews and Information Gathering

A researcher-designed questionnaire was developed for data collection. Use was made of the focus group approach or technique. This is a form of

information gathering technique where several participants are brought together to discuss a topic of mutual interest. Morgan (1988) envisages the major advantage of focus group technique as to offer the researcher a chance to observe participants engaging in an interaction that is concentrating on attitudes which are an area of interest to the researcher. The interviews with the mothers evaluated the child's adaptive behaviour and a review of the services received by the child and the family, other than the S.T.A.R.T. programme. In addition, information about the children's health status was also collected. Other types of data included the following: collection of basic socio-economic information on the family, attitude of mothers, stress coping network, social support and family function, and effects of the child's disability on family life. Information from each parent/child pair was recorded and coded (See Appendix A).

2.5 ETHICAL APPROVAL

The study was approved by the Ethics Committee of the University of Witwatersrand (Reference Number R14/49 for protocol number M931001 - M931004).

Furthermore, permission for the study was obtained from the following:

(a) Superintendent and Matron of Baragwanath Hospital

(b) Head of Speech Department of Baragwanath Hospital.

2.6. ANALYSIS OF DATA

The information and data collected were recorded and coded. Statistical analysis was performed using a SAS Statistical analysis package, involving the computation of percentages for categorical data, as well as means and standard deviations.

2.7 LIMITATIONS OF THE STUDY

2.7.1 Sample Size.

The obvious limitation is the small size of the sample. The relatively small number of infants (n=20) for both groups could contribute to the sampling error as well as the lower reliability in the statistical tests used for analyses.

2.7.2 Selection Effects.

From the initial number of 30 children with Down Syndrome identified to participate in the study to comprise the treatment group, 30% (n = 10) was lost by the time the study commenced. Withdrawal was explained to have resulted from either family relocations to areas outside the study area and/or the death of the study child. There was not enough available data regarding these families to make useful deductions as to whether they were any different from the children in the treatment group.

Of particular difficulty in this study was to match participants for age between the experimental and control groups. This difficulty arose from the fact that

the majority of mothers and/or caregivers who were referred to the Speech Therapy Department at the time of this study took up the offer to enrol in the S.T.A.R.T. programme. This left the researcher with little choice other than to take on the remaining children in the hospital records to serve as the control group. Evidently, the majority of these children's names came from older records, and were, accordingly chronologically older than those who were coming up for the first time at the time of the study.

2.7.3 Problems of securing a pure control group.

All children with Down Syndrome under the ages of three years are eligible for enrolment in an early intervention programme. At Baragwanath Hospital, these children are routinely referred, from the time of diagnosis at birth, to the Speech Therapy Department for enrolment in the S.T.A.R.T. programme. Consequently, it was not possible to undertake a random assignment of these children for this particular study. In addition it would be highly unethical and also not feasible to retain a control group for study purposes over an extended period of time without providing some services.

2.7.4 History and testing effects.

History (i.e. an individual's background) would prove to be a significant threat to the validity of the study if the sample of Down Syndrome children and their families receive other services that could account for the difference in outcome;

in addition to those provided by the S.T.A.R.T. programme. This threat was minimised in this study by the collection and analyses of data related to other services, which were received by the families and their children with Down Syndrome. The threats on testing effects were minimised by the use of a single investigator, the researcher, who was trained in the use of Griffiths Development Scales.

2.7.5 Sensitivity of Outcome Measures

Although the measures of outcome (Griffiths Development Scale) were sensitive to developmental changes, they may not have been sufficiently sensitive to detect treatment effect sizes of smaller magnitude. Furthermore, the programme could have had other positive effects on the mothers and their children, which were not ascertained in this study, for instance, the programme's effects on the relationship between parent and child.

CHAPTER 3: RESULTS

3.0 INTRODUCTION

Firstly the descriptive data and biographical data of all children with Down Syndrome, (subjects and controls) who participated in this study, will be presented. Secondly, the relevant data of the caregivers, mothers and fathers will be listed. Thereafter the results of the Griffiths Development Test done on all the children with Down Syndrome involved in the study will be given. Lastly the socio-psychological data will be presented.

3.1 AGE OF PARTICIPANTS

An attempt was made to match the children in the experimental and comparison groups by age. However, this was not entirely successful for reasons already discussed. Table V presents the age distribution of the children, who participated in this study.

TABLE V AGE DISTRIBUTION

AGE IN MONTHS	EXPERIMENT	CONTROL	STATISTICS
	(n=20)	(n=20)	
MEAN	28.4	46.9	P <0.05
STANDARD DEVIATION	19.7	19.3	
RANGE	(10.3-72.3)	(8.3-72.2)	

The mean age of the children in the control group is significantly greater than that of the experiment group, (p=<0.05)

3.2 SEX DISTRIBUTION

The sex distribution of participants in both experiment and control group is illustrated in Figure 4. There is no statistically significant difference in the distribution of sexes between the two groups $x = 1.667$, $p = 0.197$.

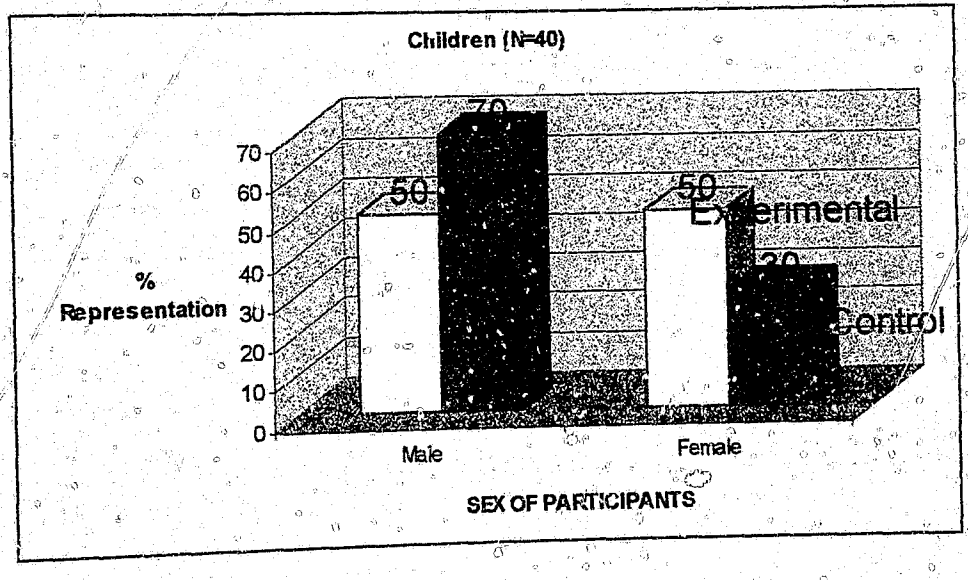


FIGURE 4: SEX DISTRIBUTION

3.3 PERINATAL DATA

3.3.1 Problems in Pregnancy

Table VI shows that there were as many problems during pregnancy in the experimental group as they were in the control group.

TABLE VI: PROBLEMS IN PREGNANCY

	EXPERIMENT	CONTROL	STATISTICS
	(n=20)	(n=20)	
YES	9	9	NS
NO	11	11	NS

Table VII gives the breakdown of the various problems encountered by mothers during pregnancy. Several mothers had more than one complaint.

TABLE VII: BREAKDOWN OF PREGNANCY PROBLEMS

LIST OF PROBLEMS ENCOUNTERED	EXPERIMENT	CONTROL	STATISTICS
	n=9	n=9	
CAESARIAN SECTION	1	2	NS
MEDICAL INDUCTION	3	2	NS
HYPERTENSION	4	3	NS
POLY HYDRAMNIOS	1	0	NS
ANAEMIA	0	3	NS
ANTEPARTUM HAEMORRHAGE	3	2	NS
DIABETES MELLITUS	3	1	NS
DECREASED FOETAL MOVEMENT	2	2	NS
BACKACHE	2	1	NS

3.3.2 BIRTH HISTORY OF THE CHILDREN

In both experimental and control groups 5 (of twenty) did not have a normal birth history. Three children in the experimental group were born after an induction (2 in the control group) and 1 child was born by Caesarian section (two in the control group).

3.3.3 GESTATIONAL AGE AT BIRTH

The gestational ages (as measured by weeks) of the two groups are illustrated in Table VIII.

TABLE VIII: GESTATIONAL AGES AT BIRTH

GESTATIONAL AGE (WEEKS)	EXPERIMENT	CONTROL	STATISTICS
	(n=20)	(n=20)	
MEAN	39.0	38.8	NS
STANDARD DEVIATION	1.5	1.7	
RANGE	(4-41)	(31-40)	

There were no significant statistical differences between the two groups.

3.3.4 NEONATAL HISTORY (N=40)

There was no statistically significant difference in the percentage of problems in the neonatal period between the experiment group (7 out of 20) as against the control group (10 out of 20). The problems encountered in neonates are given in Table IX:

TABLE IX: PROBLEMS IN THE NEONATAL PERIOD

PROBLEMS IN NEONATAL PERIOD	EXPERIMENTAL n=20	CONTROL n=20
NEONATAL JAUNDICE	2	4
CARDIAC DEFECTS	4	4
VOMITING	1	2

3.4 THE AGE OF ONSET OF DEVELOPMENTAL MILESTONES

Table X shows the distribution of mean-ages of onset of developmental milestones of the two groups. In every case the Down Syndrome children in the experimental group achieved their milestones significantly earlier than those in the control groups.

TABLE X: THE DISTRIBUTION OF AGE OF ONSET OF DEVELOPMENTAL MILESTONES (N=40)

AGE (MONTHS)	EXPERIMENTAL GROUP	CONTROL GROUP	p-VALUE
A: SITTING	(n=17)	(n=19)	0.018
Mean	7.5	11.1	
SD	3.8	3.1	
Range	(6-14)	(4-24)	
B: WALKING	(n=11)	(n=17)	0.006
Mean	12.3	21.2	
SD	12.1	13.3	
Range	(13-35)	(12-48)	
C: FIRST WORD	(n-12)	(n=17)	0.0001
Mean	10.9	31.1	
SD	10.9	17.0	
Range	(9-30)	(30-70)	

3.5 CAREGIVER REPORT ON ASSOCIATED HEALTH PROBLEMS OF THE CHILDREN WITH DOWN SYNDROME

Earlier cardiac complications, a great percentage of which were associated with ventriculo-septal defects, were reported to the same degree (20%) in both experimental and control groups. Cardiac surgery was performed in twenty percent (20%) of the experimental group (n=4) and fifteen (15%)

percent of control group (n=3) as shown in Table XI.

TABLE XI: PROBLEMS ASSOCIATED WITH DOWN SYNDROME

	EXPERIMENT n=20	CONTROL n=20
EYE	0	1
HEART	4	4
CHEST	4	0
GASTRO-INTESTINAL	1	0
FAILURE TO THRIVE	0	1
CARDIAC SURGERY	4	3

3.6 CAREGIVER REPORT ON PRESENT COMPLAINTS

Coughing or chest problems were the most frequently reported current symptoms (Table XII). Except for chest related problems, medical problems were diverse and fairly evenly distributed among the experimental and control groups as shown in Table XII.

TABLE XII CAREGIVER REPORT ON PRESENT COMPLAINTS (N=40)

	EXPERIMENTAL (n=20)	CONTROL (n=20)
PROTRUDING TONGUE	0	1
DENTAL CARRIES	1	1
CONSTIPATION	1	1
SQUINT	1	1
COUGH	3	1
CARDIAC	0	1

These observations suggest that of the 4 children in the experimental group who had early cardiac surgery, none was reported by the caregivers to have any further complaints in this regard. For the control group, of which three had cardiac surgery, it would appear that two were well, while in one had some cardiac complaints were noted.

3.7 DESCRIPTIVE DATA OF CAREGIVERS COMPARED WITH THAT OF THE MOTHERS AND FATHERS

3.7.1 Mean Ages

Whilst there was no significant difference between the ages of the experimental and control groups, in the maternal (35.2years versus. 34.4years) and paternal (40.4years versus 34.7years) mean ages, the caregivers in the control group, were numerically much older (mean age 46.6 years) than the experimental caregiver group (mean age 34.2 years) (Figure 5.). There was no statistically significant difference between groups. It may also be pointed out, as stated later on in section 4.2.2b, that there were significantly more mothers as caregivers in the treatment group (70%) than in the control group (40%). It is possible that the relatively younger mothers were more predisposed towards taking part in the S.T.A.R.T. programme than the older caregivers.

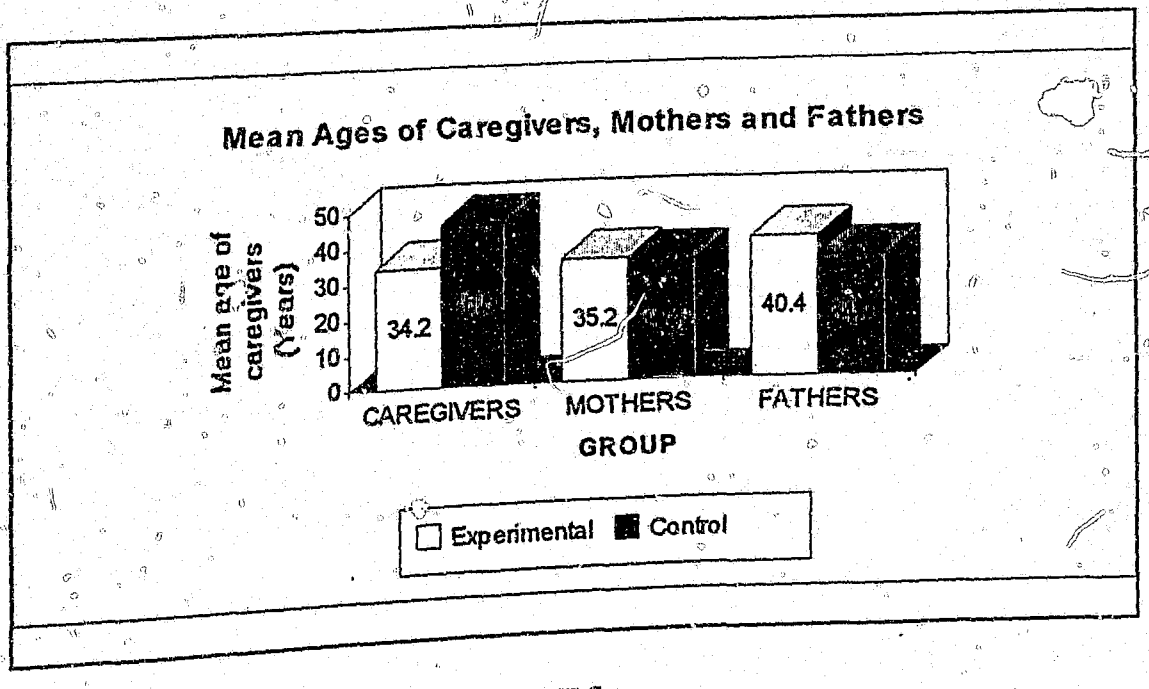


FIGURE 5: MEAN AGES OF CAREGIVERS

3.7.2 EDUCATION OF CAREGIVERS, MOTHERS AND FATHERS

A comparison of educational levels of participant caregiver, mothers and fathers for both experimental and control groups as shown in Table XIII.

TABLE XIII: EDUCATIONAL HISTORY OF CAREGIVERS, MOTHERS AND FATHERS

EDUCATION	% CAREGIVER		%MOTHERS		%FATHERS	
	EXP	CONT	EXP	CONT	EXP	CONT
NO SCHOOLING	0	45 ($p<0.05$)	0	5	10	25
STANDARD 1-5	30	25	30	20	35	10
STANDARD 6-8	30	15	30	30	20	40
STANDARD 9-10	35	10	35	30	30	10
TERTIARY	5	5	5	15	5	15

All differences were not significant at $p<.05$ except for the number of uneducated caregivers in the control group versus the experimental group.

3.7.3 COMPARISON OF OCCUPATION OF CAREGIVERS, MOTHERS AND FATHERS

A similar percentage of caregivers and mothers in the experimental group was unemployed (45%) compared to the control group where the percentages were 15% for the caregivers and 20% for the mothers (Table XIV). 40% of control group caregivers were students. Whilst 40% of fathers in the experimental group were skilled only 15% of the control group were skilled.

TABLE XIV: OCCUPATIONAL COMPARISONS OF CAREGIVERS

	% CAREGIVERS			% MOTHERS			% FATHERS		
	EX	CON	STA	EX	CON	STA	EX	CON	STA
NEVER WORKED	10	5	NS	10	10	NS	5	5	NS
UNEMPLOYED	45	15	P<.05	45	20	p<.05	10	25	NS
UNSKILLED	25	25	NS	25	30	NS	30	40	NS
SKILLED	15	10	NS	15	10	NS	40	15	p<.05
SEMI PROFESSIONAL	5	5	NS	5	15	NS	10	10	NS
PROFESSIONAL	0	0	NS	0	15	NS	0	0	NS
STUDENT	0	40	P<.05	0	0	NS	5	5	NS

80% of fathers in the experimental group were employed compared to 65% in the comparison group. Further analyses indicated that all the mothers involved in caregiving both in the experimental (70%) and control (40%) groups did so in varying degrees under varying circumstances, for example, in employment and study commitments.

3.7.4 MARITAL STATUS

There was no significant difference in the marital status of the two groups, with experimental group (15 married, 5 single) and control group (12 married and 8 single)

3.8 MEASURES OF SOCIO-ECONOMIC STATUS AND OTHER FAMILY CIRCUMSTANCES

Figures 6 to 9 illustrate profiles of the socio-economic circumstances of the participating families on a number of indicators. These figures are presented in a manner which facilitates comparisons between the experimental and control/comparison groups.

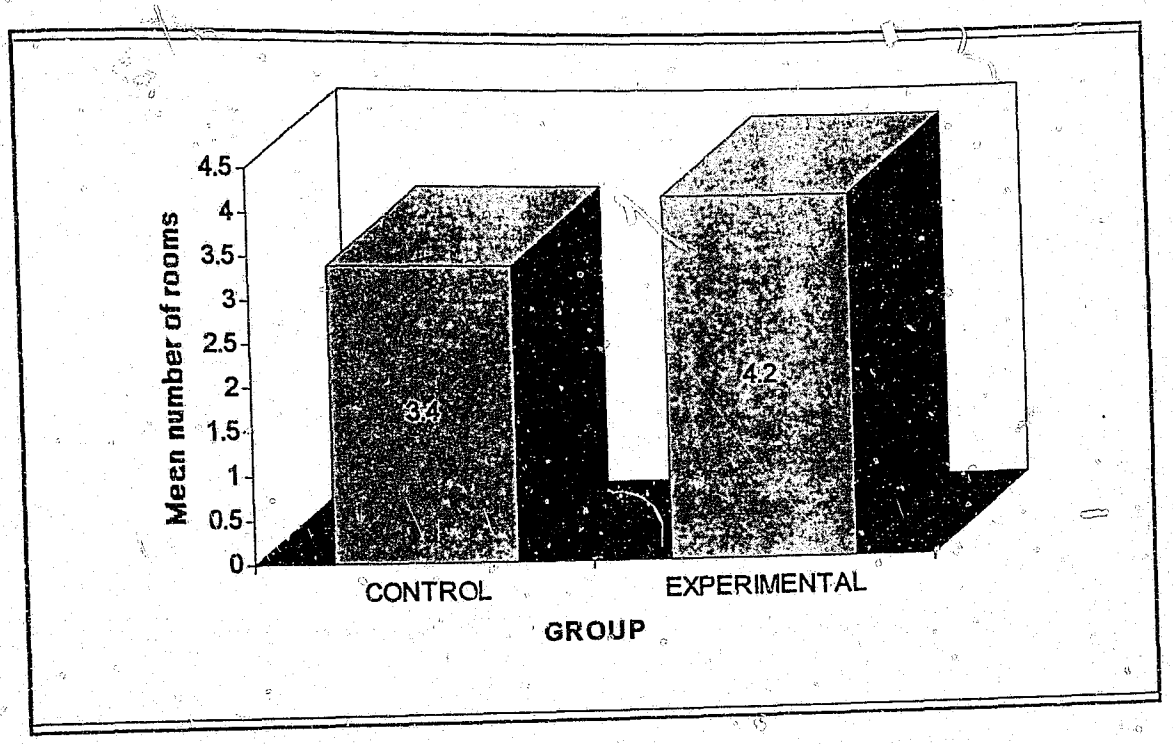


FIGURE 6: MEAN NUMBER OF ROOMS IN A HOUSE

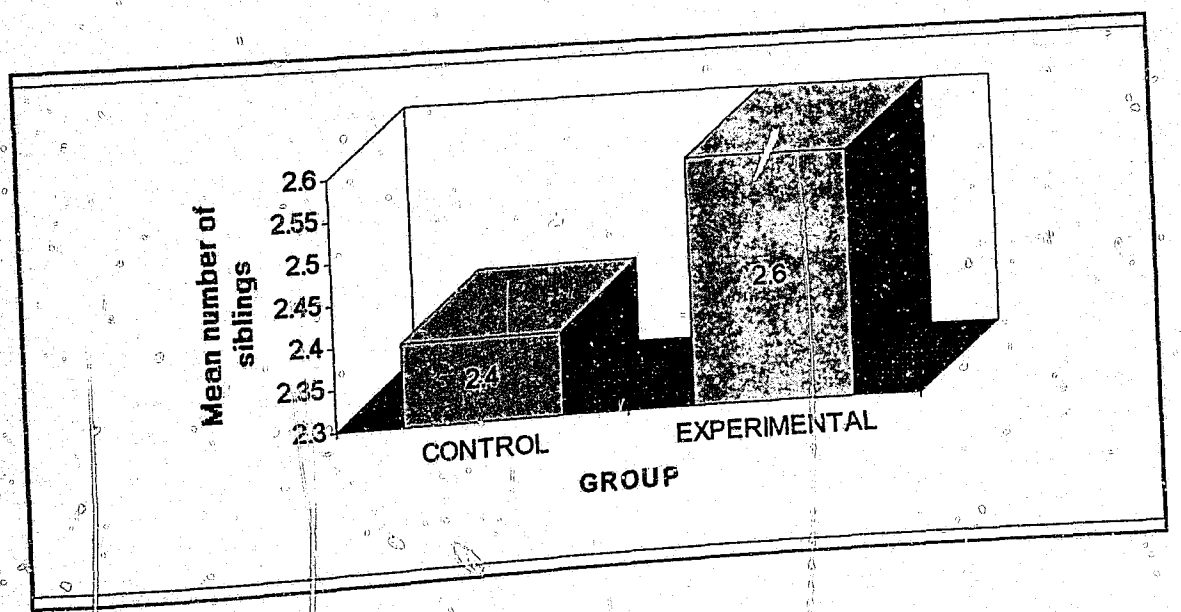


FIGURE 7: MEAN NUMBER OF SIBLINGS

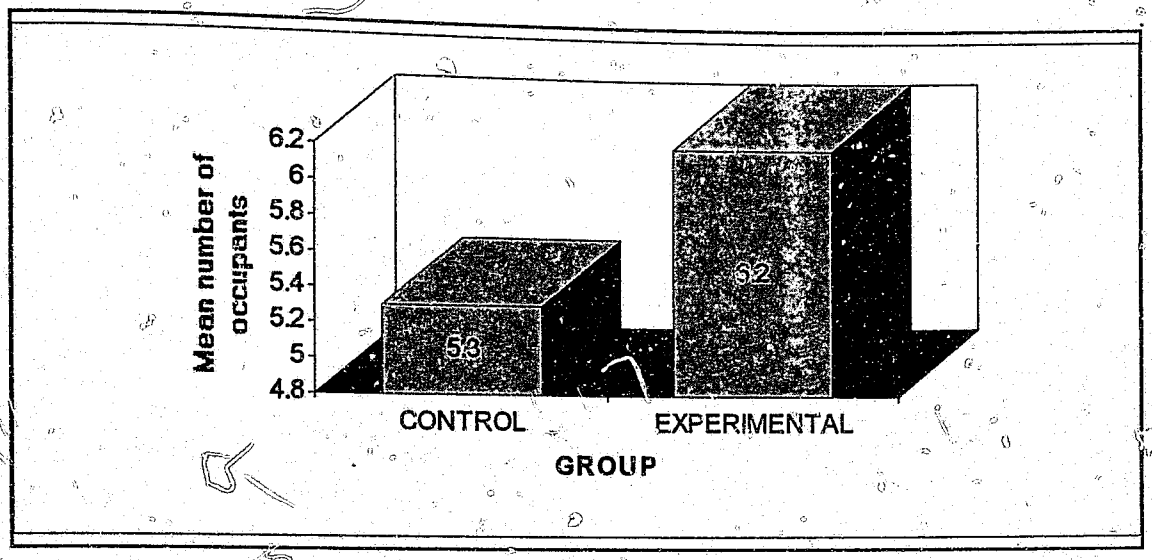


FIGURE 8: MEAN NUMBER OF OCCUPANTS

In figures 6,7 and 8 it is clearly shown that there was no significant difference between the experimental as opposed to the comparison group in the areas of number of rooms, mean number of siblings and mean number of occupants.

Figure 9 below shows a similar profile of paternal support between the experiment and control groups as reported by the caregivers.

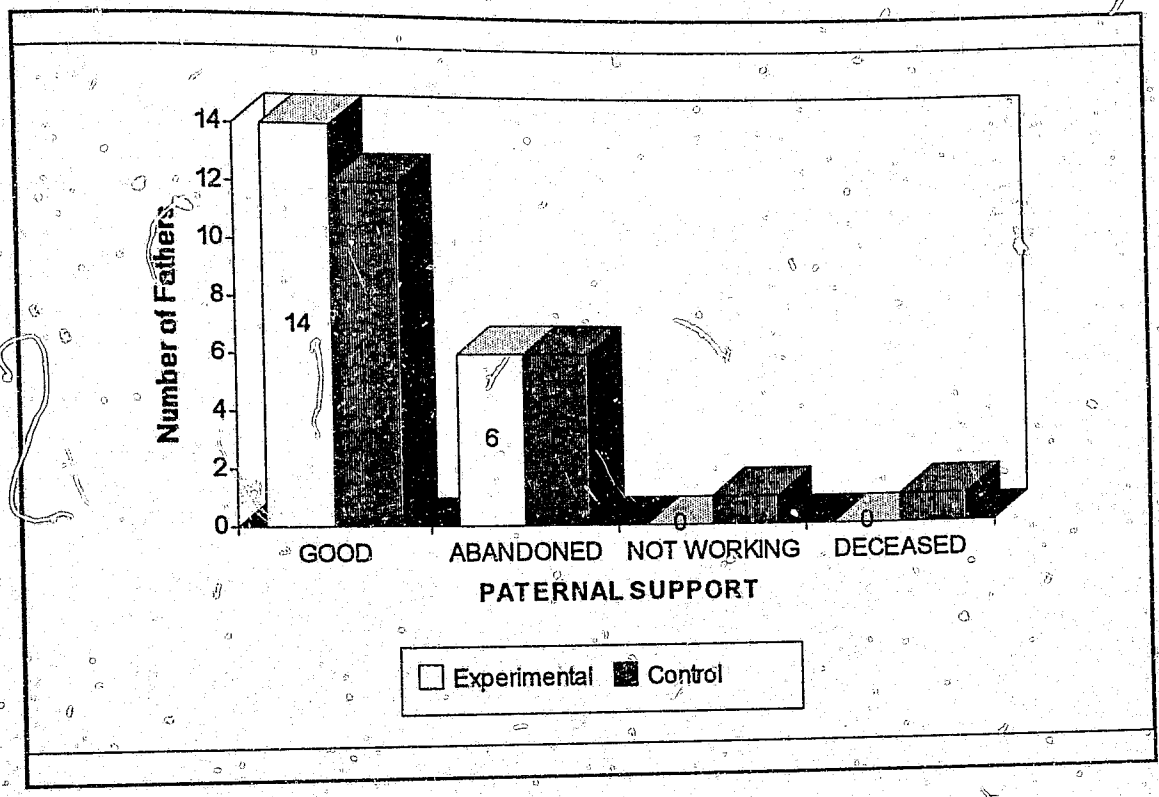


FIGURE 9: PATERNAL SUPPORT

3.9 BIRTH POSITION OF AFFECTED CHILD

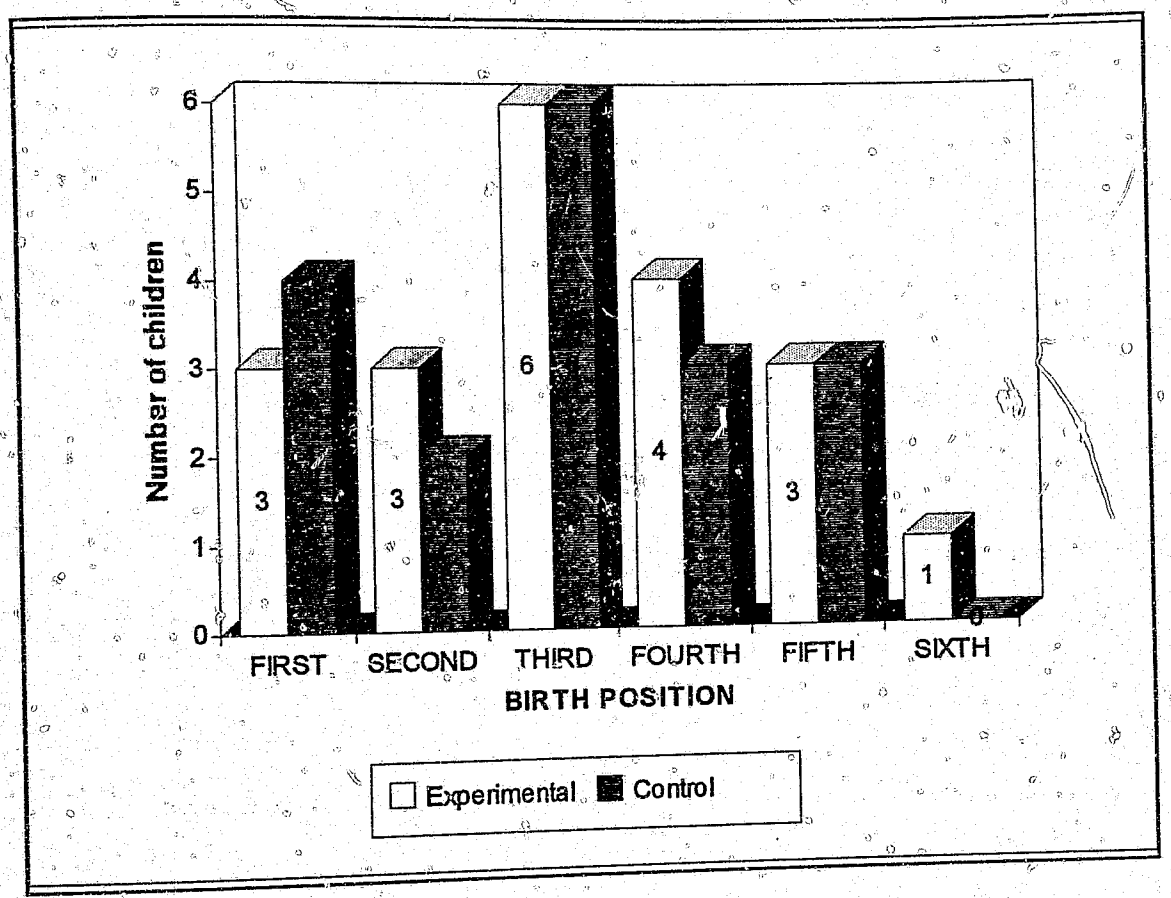


FIGURE 10 BIRTH POSITION OF AFFECTED CHILDREN

The pattern of birth order between the two groups is similar.

3.10 FAMILY HISTORY OF DOWN SYNDROME

Although none of the siblings of the affected children had been diagnosed to have Down Syndrome, 25% in the experimental group and 35% in the control group (Table XV) had a positive family history.

TABLE XV: FAMILY HISTORY OF DOWN SYNDROME

	EXPERIMENT	CONTROL	STATISTICS
MATERNAL	4	1	NS
PATERNAL	1	6	$p<0.05$
SIBLINGS	0	0	NS

3.11 GRIFFITHS DEVELOPMENT SCALES

TABLE XVI: RESULTS

		GROUP		p-value	90% CI ratio
		S.T.A.R.T	CONTROL		S.T.A.R.T to CONTROL
PARAMETERS	STATISTICS			t-test	
LOCOMOTOR	X	50.1	53.4	0.59	0.28≤ ≤1.16
	SD	21.39	16.9		
	N	20	20		
PERSONAL SOCIAL	X	50.7	43.6	0.21	0.08≤ ≤1.41
	SD	21.45	12.35		
	N	20	20		
HEARING AND SPEECH	X	46.0	35.8	0.05	0.001≤ ≤1.56
	SD	19.17	11.6		
	N	20	20		
EYE-HAND CO-ORDINATION	X	42.9	37.6	0.29	0.88≤ ≤1.40
	SD	18.92	12.04		
	N	20	20		
PERFORMANCE	X	41.10	34.4	0.15	0.94≤ ≤1.45
	SD	15.54	13.09		
	N	20	20		
PRACTICAL REASONING	X	43.3	52.2	0.09	0.63≤ ≤1.21
	SD	7.6	18.81		
	N	8	8		
DEVELOPMENT QUOTIENT	X	45.6	45.9	0.69	0.85≤ ≤1.23
	SD	14.81	12.3		
	N	20	20		

The Griffiths Development Scales results are summarized in Table XVI. The mean Development Quotient (DQ) for the experimental group was 45.61 (SD=14.8),

compared to that of the control group which was 45.85 (SD=12.3). Differences between the two groups did not reach a significant difference except for the speech pr[illegible] there is a significant difference at $p = 0.05$ In the practical reasoning profi[illegible]ntrol group performed better than the experimental group at a $p = 0.097$, which was also approaching significance.

3.12 PSYCHO-SOCIAL DATA

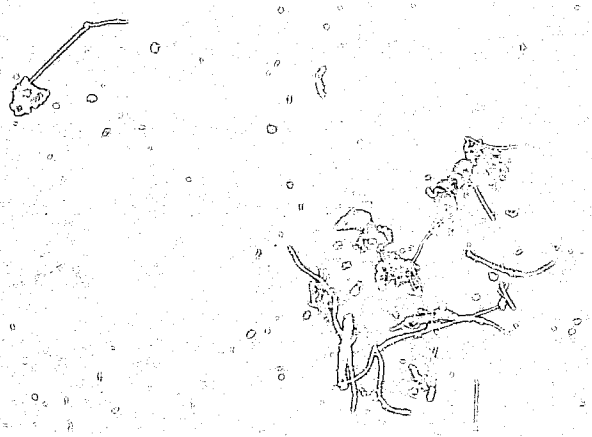
TABLE XVII: SUPPORT SYSTEMS OF FAMILIES OF CHILDREN WITH DOWN SYNDROME

	EXPERIMENTAL	CONTROL	STATISTICS
THERAPIST (START)	20	3	$p < 0.05$
SOCIETIES	7	13	NS
OTHERS	3	2	NS

Table XVII indicates that 100% of the participants in the experimental group depended on the S.T.A.R.T. therapist for support whereas the majority of the control group depended on other resources within the communities.

3.13 THE CAREGIVER ATTITUDE TO S.T.A.R.T. PROGRAMME

Ninety-five percent of caregivers in the experimental group claimed to have had positive results from participating in the START programme. None of the members of the control group reported any previous exposure of the children to the S.T.A.R.T. programme. Thirty five percent of the control group had participated in alternate intervention programmes; 65% of these had physiotherapy; 14% were receiving special schooling and another 14% had speech therapy only.



CHAPTER 4: DISCUSSION

4.0 INTRODUCTION

In order to interpret the results of this study, a few general comments have to be made regarding studies that evaluate the efficacy of early intervention in general and in children with Down Syndrome in particular.

One major obstacle in the assessment of the progress of handicapped infants is the scarcity of feasible research designs (Bricker et al, 1981). Many studies cannot implement a random control trial due to the ethical concerns surrounding the withholding of services from infants assigned to control groups. Furthermore, the rights of children with handicaps in many countries mandate provision of educational services and programmes, which eliminates possible subjects as controls for research of this nature. Many studies therefore have to rely on an *ex post facto* design, as is the case in this study.

Although every effort can be made to eliminate major differences between the experimental and control groups (and the utilization of waiting lists for the 'control' group in clinical research is often practised), there are no guaranties that these two groups may not differ significantly on variables such as age, sex distribution, socio-economic status and so on. Since early intervention studies should be done to ascertain their efficacy, and *post facto* designs are more likely to be utilised, the

conviction with which differences in outcome measures can be ascribed to the programme, will be limited by the number of co-variables that are dissimilar between the two groups. In the interpretation of the outcome measures, more than one proposal for the di[illegible]s may have to be entertained.

Furthermore, it would appear from previous studies that the ability to demonstrate the efficacy of an early intervention programme may depend on the intervention programme itself, length of intervention, the number of children assessed, the sensitivity of the outcome measures and the physical health of the children (Bricker et al, 1981; Shea, 1993).

In this study, the effects of the S.T.A.R.T. programme in the development of infants and children with Down Syndrome were evaluated using the Griffiths Development Scales. In addition, the children's abilities in the following domains were compared: locomotor, hearing-speech, hand-eye co-ordination, performance and personal reasoning. Furthermore, some aspects of impact of the programme on the parents and care givers was also assessed. The results will now be discussed against the backdrop of the constraints of this kind of research in general, and the limitations of this study in particular.

4.1 GRIFFITH'S DEVELOPMENT SCALES

4.1.1 Hearing and speech

This was the only domain where there was a significant difference between groups, the experimental group performing significantly better than the control group. Problems of speech and language development in the children with Down Syndrome in the domain of expressive or productive communication are well described in literature (Collacott, 1993; Dykens et al (1994); Miller, 1988). These were particularly obvious in the specific areas of babbling (Dodd, 1972; Steffens, 1992) and linguistic imitation (Clark, 1977). Dykens et al (1994) further pointed that their weaknesses became more pronounced as children with Down Syndrome became older. Articulation problems were also found to be contributing factor (Zisk & Beiler, 1967). The period between 18-24 months is characterized by a plateau and only at 36 months do they start to use a few words spontaneously.

In this study, in the speech and hearing domain, the performance of the treatment group was significantly better than that of the comparison group ($p<0.05$). The observed results (treatment group DQ[SD] = 46(19.2) and comparison group DQ[SD] = 35.8(11.6)) are consistent with the findings from the previous controlled studies where intervention of speech and hearing in children with Down Syndrome were investigated (Bidder, Bryant & Gray, 1975; Tannock, Girolametto & Siegel, 1992). Dykens et al (1994) further

pointed out that communication weakness in children with Down Syndrome may be observed in relation with other domains of adaptive behaviour such as daily living and socialization skills. Therefore, the facilitation of language development in the treatment group is an important finding because of its possible significance for general development, in the development of communication and acquisition of self-sufficiency. The development of language was of great help to the mothers and caregivers in the present study, as they could understand their children's speech better and so were better able to communicate with their children during the various activities of daily living.

4.1.2 Personal-Social Domain

The activities of personal- social help skills were described in Appendix C: and the rate is shown to fall below that of normally developing children. The small but non-significant gain in the personal-social and performance profiles in the treatment group which is demonstrated in the present study is unlikely to be attributable to a "placebo -effect" only, because the gains measured do correspond to areas emphasized in the S.T.A.R.T. programmes activities of daily living (Evans, unpublished data). Similar reports were given from earlier studies by Bidder et al (1975)

4.1.3 Locomotor Domain

In the domain of locomotor development, there was no significant difference in the observed behaviour between the treatment and comparison groups. Early motor deficiencies in the infants with Down Syndrome are well described (Rast

& Harris, 1985). They are obvious as early as six months of life (Carr, 1993) and the evolution of postural reaction is directly related to acquisition of early milestones. Furthermore, previous studies have shown that children with Down Syndrome have difficulty in eye-hand co-ordination, balance, laterality and visual motor activity (Rast & Harris, 1985; Shumway-Cook & Woollacott, 1985).

The apparent lack of improvement in the locomotor profile in the study could be attributed to the effect of the advanced mean age of the control group (cohort effect). Brinkworth (1972) as quoted by Connolly (1993), reported that this effect may be due to a predetermined limited biological maturation in children with Down Syndrome, which may prohibit any advancement of the rate of development after a certain age. Balance, visual and motor activity continue to be problem areas for children with Down Syndrome. Accordingly, early intervention should emphasize therapeutic intervention in these areas so as to decrease functional deficit (Connolly et al, 1993; LaVeck & LaVeck, (1977)).

4.1.4. Practical Reasoning Domain

In the practical reasoning domain, the performance of the treatment group is superseded by that of the comparison group, to a degree that approaches significance (See Table XVI). This sub-scale deals with arithmetic, comprehension and problem solving. The maturation factor due to age may also be a significant contributor to this observation.

4.2 CONFOUNDING VARIABLE IN THIS STUDY

Two groups of confounding variables need further scrutiny. They relate to: (a) the subjects of experimental vs the control group, and (b) the caregivers/parents of both groups.

4.2.1. The subjects

It was found that the subjects in the experimental and control groups were similar as far as the following variables were concerned: -

(a) Sex distribution.

(b) Problems during pregnancy and birth.

(c) Neonatal history.

(d) Disease profile

(e) Birth position of the affected child.

The major confounding variable was the significant age difference between the two groups. The experimental mean age (SD) of the experimental group being 28.4 (19.7) months and that of the control group 46.9 (19.3) months.

($p<0.05$). Yet the ranges were similar (10.6-72.3) and (8.3-72.2) respectively (Table V). Although every effort was made to recruit subjects in both groups of similar age (and they all fulfilled the inclusion criteria for age), this is one of the limitations of ex post facto research, that the groups may not be similar for all variables. Unfortunately this is an important variable, as many conclusions regarding development would depend on the age of the subjects.

Despite the fact that the experimental group was significantly younger, on average, they achieved all the milestones as measured by age at sitting, walking and first words significantly sooner than their older peers in the control group (Table X). This could be a reflection of the efficacy of the S.T.A.R.T. training programme, or be due to the fact that for some reason the control group was, on average, more mentally retarded. This is not borne out by the DQ on the Griffiths' Scales, which was nearly identical for both groups, 45.6 (14.8) and 45.9 (12.3) respectively (Table XV1)

It is particularly the improved rate of language acquisition that is of interest. The experimental group uttered their first words at a mean age of 9.1 months, where as for the comparison group the age was given as 31.1 months. These findings are similar to a study by Connolly et al (1980) where the mean age for the utterance of first words was found to be 10.6 months and 24.3 months for the experimental and control group respectively. Again it can be postulated that the S.T.A.R.T. programme is effective for the acquisition of language or it

may simply reflect the differences in the social background and quality of care giving of these two groups of children with Down Syndrome.

4.2.2 The Caregivers/parents

When comparing the experimental group with the control group the following important differences emerged:

(a) The caregivers of the experimental group were younger than the control group (34.2 years versus 46.6 years).

(b) 70% of the caregivers in the experimental group were the mothers themselves (compared to 40% in the control groups)

(c) There were statistically significantly more uneducated caregivers in the control group than the experimental group (45 versus 0, $p<0.05$).

(d) 40% of the fathers in the experimental group were working in skilled occupations, versus 15% in the control group ($p<0.05$).

(e) 40% of the control group's caregivers were still students, which could mean there was lack of insight on their part, as they were still young. They also could not give undivided attention to caregiving. None of the experimental group's caregivers were still at school.

There were no significant differences between the two groups as far as the following were concerned:

(a) Marital status

(b) Paternal and maternal age.

(c) Socio-economic parameters.

(d) Mean number of rooms in house

(e) Mean number of siblings

(f) Mean number of occupants in house

(g) Paternal support.

Child development is dependant on the resources available to a child via his family and the community, which form the basic equipment for competence (Edgar, 1995; Ruskin et al, 1994). This is even more important in the development of children with handicaps. The families and home environment have become important variables in the study of child development (Bronfenbrenner, 1986, Richter and Grieve, 1991). Factors that have been identified to have a negative impact on child development are low paternal education (Bester, 1994), poor housing (Goduke, Poole & Aokati-Phenise, 1997) and overcrowding (Gordon et al, 1992; Graham, 1971). Neighbourhood factors also play a role in cognitive outcomes (Chase-Lansdale & Gordon, 1996). For most of these variables there were not significant differences between the experimental and control groups in this study, except that the fathers were more skilled and more likely to be employed in the experimental groups and the mothers better educated. A major factor that has to be taken into consideration when studying child development is in whose care and in whose company children spend their time (Harkness, 1992). In this study, the differences on the descriptive data of the caregivers of the two groups, emerge as an important confounding variable.

Not only is the control group cared for by persons who were older and less educated, they were also less likely to be the mother. Research has shown that parental involvement and motivation to assist their disabled children was the most important factor in the whole spectrum of the rehabilitation of children (O'Toole, 1989; Piper et al, 1980; Ramey, Bryant & Wasik, 1992). More than 50% of the mothers of the experimental group were not employed and could therefore attend to their children, compared to 20% in the control group. The mothers in the experimental group are thus more likely to be available to their infants and more likely to participate in an intervention programme. As their husbands enjoyed better employment there may also have been less pressure on them to be employed as well.

As the mothers in the experimental group were also better educated (70% were educated above standard six), there may have been a greater chance that they would request that their children be given an opportunity in an early intervention programme, such as S.T.A.R.T. (Sharav, Collins and Shlomo, 1985).

It could therefore be argued that it was the micro-environment of the experimental group that was conducive to improved development, rather than

the intervention programme per se. Improved language ability could therefore be the direct result of improved stimulation and caring by caregivers who were superior because they were the mothers, better educated, came from an improved socio-economic environment and were younger. It is still possible that they could have benefited from the S.T.A.R.T. programme, as it would have given them the opportunity to structure their interaction with their Down Syndrome children in such a way as to make their stimulation and interaction more effective.

4.3 S.T.A.R.T. AS AN EARLY INTERVENTION PROGRAMME

Although research on effectiveness of early intervention programmes is still controversial, there is important evidence that would suggest positive effects of such intervention on children with Down Syndrome (Cunningham, 1986; Shonkhoff & Hauser-Cramp, 1992). There are two major benefits of early intervention programmes, those experienced by the parents and those experienced by the child with special needs. As far as the parents are concerned, the negative effects of having a child with a disability, is well described in literature. Such a parent may have feelings of isolation, depression and helplessness (Solarsh, 1986; Quine et Pahl, 1987). Participation in programmes helped to reduce the feelings of helplessness and empowered the parent to work effectively with the child. This is made possible by the encouragement provided by the professionals engaged in such a programme

and the support given by other parents. This gives an opportunity for the parents to learn of the child's strengths and weaknesses under the guidance by professionals. They learn to be advocates for their children at an early stage.

The S.T.A.R.T. programme is intended to support both parents and children with special needs. It is locally developed and so grounded in common values and principles. Katz (1995) studied perceived effects on parents who had regular contact with the S.T.A.R.T. programme and concluded that the most helpful aspect included support, positive reinforcement and motivation to continue. Katz also found that the coping capacity of mothers improved 100% for those who were involved with the S.T.A.R.T. programme, and 70% of these mothers felt their expectations were met.

In a meta-analysis review by Simeonsson et al (1982), it was concluded that, although statistical significance was not attained, children with disabilities involved in early intervention programmes (EIP), seemed to make better progress than those not included in such programmes. Such children involved in the programme often made progress in areas not measured in the studies and also areas specific to the child. The S.T.A.R.T. programme may have had other positive effects for the mother, the child and the family that were not identified and measured, for example, the affective relationship between mother and child, or reducing stress in the family (which would not have been

identified by the Griffiths' Development Scale). The child's behaviour, or style of response may have improved, but not documented. There may be a maintenance of a certain level of development, or prevention of regression which may also be reflective of success, but not recorded. In addition, there may be no improvement in the developmental domains, but improvement in the management areas, for example, in seizure control or in feeding. The lack of empirical evidence or support of effectiveness may be a function of the nature of the measurements, as well as the timing and duration of the intervention.

CHAPTER 5: CONCLUSION

The aim of this study was to investigate the benefit of the S.T.A.R.T. programme on the development of children with Down Syndrome between the ages of 6 months to 6 years, using the Griffiths' Development Scales. The results of the study, show that the group of children who attended the S.T.A.R.T. programme performed better than those who did not attend, in the areas of acquisition of milestones; hearing and speech; social and personal developmental domains of the Griffiths test. However, despite the positive findings mentioned above, the anticipated treatment effect of acquisition of a significant developmental change, overall, in favour of the treatment group was not found. As previously emphasized, the majority of the control group participated in other pre-school and other community activities, although none had received specific S.T.A.R.T. programme instructions. Therefore, the improvements in the control group most likely reflect the cumulative effects of maturation and general stimulation. The positive findings in early acquisition of milestones, better performance in speech and hearing domain, and social and personal domains cannot be wholly attributed to the S.T.A.R.T programme but may also be due, in part, to the effects of the positive caregiver environment bestowed by the young educated caregivers in the experiment group. The majority of these caregivers were the mothers who had no need to work as they were adequately financially supported by their working husbands.

However, the S.T.A.R.T. programme may still have an important role to play as it provided the structure, the system and guidelines for the intervention.

The results of this study should not be interpreted as evidence that the S.T.A.R.T. programme cannot be effective in facilitating development in children with Down Syndrome, but that its adoption for such a purpose needs closer scrutiny.

There is need for random control studies which should focus on the effectiveness of the S.T.A.R.T. programme in a variety of circumstances, both in an attempt to replicate the findings reported in this study, as well as extend them. Further it also appears necessary to examine available outcome measures such as Griffiths' Development Scales, with a view to evolving more sensitive instruments which could provide both quantitative and qualitative data. Such data would greatly enrich the process of measuring the advantage (or lack thereof) of various developmental programmes, both currently available and those yet to be developed and should also enhance the efforts to document that early intervention is not only humane but effective (Simeonsson, 1982).

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