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DEPARTMENT OF CLINICAL SCIENCES IN MALMÖ
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**CLEFT LIP AND PALATE IN
ADOLESCENCE
IDENTIFYING VARIABLES RELATING
TO PSYCHOSOCIAL CONCERNS**

MARTIN PERSSON

MALMÖ
2007

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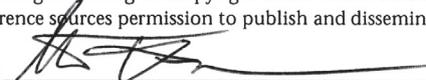
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take place in the lecture hall
at the Department of Medicine, entrance 35,
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| Abstract <p>In study I, 55 adolescents with cleft lip and palate and 31 adolescents without cleft filled out two questionnaires: the Tennessee Self-Concept Scale to assess self-concept, and the Eysenck Personality Questionnaire Inventory to assess the degree of introversion. In studies II & III, data regarding physical characteristic and general intellectual capacity were obtained from the Swedish National Service Enrolment Register for the years 1991-97. Data concerned 335 men with cleft lip with or without cleft palate and 88 with cleft palate. They were compared with a control group of 272 879 men without cleft. In study IV, data regarding academic achievement at the time of leaving compulsory education were obtained by linking the Swedish Medical Birth Register for the years 1973-86 with the Swedish School Grade Register. Data concerned 651 individuals with cleft lip, 830 with cleft lip and palate, and 511 with cleft palate. They were compared with a control group consisting of 1 249 404 individuals without cleft. Adolescents with cleft seem to have a normal to high self-concept without an accentuated degree of introversion (I). A cleft influences body dimensions, particularly in the cleft palate group where weight, stature and muscle strength (II) showed lower values, this may be correlated to their reduced achievement in physical education at school (IV). The general intellectual capacity (III) as well as the academic achievement (IV) were both mostly adequate for adolescents with cleft lip with/without cleft palate, whereas adolescents with cleft palate performed somewhat worse on average.</p> | | |
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MARTIN PERSSON

MALMÖ
2007

TO
NIKOLAI, SOFIA, ALEXANDER AND HENRY

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ABBREVIATIONS

| | |
|---------|---|
| ANOVA: | Analysis Of Variance |
| BMI: | Body Mass Index |
| EPQ-I: | Eysenck Personality Questionnaire Inventory |
| GIC: | General Intellectual Capacity |
| IQ: | Intelligence Quotient |
| MBR: | Medical Birth Register |
| N: | Muscular Strength (Newton) |
| OR: | Odds Ratio |
| PD: | Pass with Distinction |
| PE: | Pass with Excellence |
| RCM: | Registry of Congenital Malformations |
| TSCS: | Tennessee Self Concept Scale |
| SEB: | Swedish Enlistment Battery |
| 95% CI: | 95% confidence intervals |

INTRODUCTION

It is postulated that individuals with cleft lip and palate may be at risk to experience psychosocial problem based upon their congenital malformation. Since cleft is one of the more common congenital malformations it is of importance to address this postulation. The incidence rate in Sweden is about 1.8/1000 newborns [1, 2], however the global distribution of cleft varies with geographic location, ethnic group and socioeconomic conditions [3, 4]. In 2005 in Sweden, 96 children were born with cleft lip with or without cleft palate and 58 children with isolated cleft palate [5]. All of them will be followed up into late adolescence and receive habilitation by a multidisciplinary team comprising representatives from plastic surgery, orthodontics, oto-rhino-laryngology, phoniatrics, speech therapy and social work.

The issue of psychosocial aspects in relation to cleft is though not addressed to the same extent as results regarding maxillofacial growth, speech, hearing and facial appearance. It is internationally recognized that more focus needs to be addressed to psychological and quality-of-life measures [4]. The psychological aspects in relation to cleft have been examined for decades with an assortment of outcomes and conclusions with various methodological approaches. Hunt and coworkers (2005) conducted a systematic review of the psychosocial effects of cleft lip and palate. They looked at 652 abstract from which they determined that 64 articles met the criteria to be included in their review. However it was not possible to conduct a meta-analysis of the studies included secondary due to the large variations in study design and outcome measures [6]. Hence it becomes intricate to formulate conclusions based upon the methodological limitations, their review indicate that there are some limited evidence of psychosocial problems in certain areas, but the overall functioning appears to be adequate for individuals with cleft [6].

This raises a fundamental question that is worth considering; is it right to assume that an individual with cleft will have an increased risk of psychosocial problems completely based upon the cleft *per se*. Many psychosocial studies follow this notion, including the first article in this thesis, which basically compare how individuals with cleft function in relation to the general population in a specified area or concept. This arbitrary notion needs to be questioned since it does not take into account other variables or determinants that has a crucial influence on the psychosocial wellbeing [7]. It is then imperative to identify what kind of variables are influenced by cleft, if any, so they can be added into the equation when analyzing the psychosocial aspects for individuals with cleft. The other articles in this thesis focus upon identifying potential variables that could have an impact on the psychological or quality-of-life measure.

Self concept and introversion

When it comes to self-concept, individuals with cleft lip and palate seem to perceive themselves differently depending on age. Studies on children have shown diverging results with both a lowered self-concept [8, 9] and a medium to high self-concept [10]. Compared with children, adolescents with cleft lip and palate more consistently show an average to high self-concept [10, 11].

The degree of inhibition is significantly correlated to the degree of psychological maladjustment, and may thus depend on the individuals with cleft lip and palate behaviour and social skill [11]. These qualities are at least partly determined by the acceptance an individual with cleft lip and palate will experience in the environment. However, people in general may be hesitant or even negative towards individuals who do not display normal human features or behaviours. Regarding the operated individual with cleft lip and palate, the appearance is actually considered less friendly and popular and not so intelligent and attractive compared with the normalised appearance achieved by retouched photographs [12]. It is therefore not surprising that an individual with cleft lip and palate often is concerned about facial appearance. In childhood and adolescence, this correlates with an increased social inversion and may also be accompanied by poor self-concept and anxiety [13, 14].

Physical characteristics

It has been thought that cleft lip and palate and cleft palate with their attendant structural malformations and related functional problems may affect general physical development. Possible explanations for such physical development are ontogenetic conditions [15], inadequate nutrition [16, 17] or deleterious effects of reconstructive surgery [18]. A genetic disposition to a cleft may therefore be associated with genes that code for general physical development individually.

Studies have been conducted on patients with cleft lip and palate and cleft palate to find out if the structural malformation contributes to significantly lower body weight than the normal population. A number of studies indicates that there is no appreciable impact on body weight [19-21], but some research has indicated that if a patient has unilateral cleft lip and palate, or isolated cleft palate, and is male, then there is a possibility of lower body weight [22]. In support of the last notion, some studies indicate that there is a significant influence on birth weight [23, 24]. As far as stature is concerned some researchers think that cleft lip and palate and cleft palate may be associated with an increased risk of impeded growth [20, 22, 23, 25, 26] though other reports do not [16, 19, 21, 27].

There is therefore no consensus whether weight and stature are affected by the presence of a cleft. Weight and stature are themselves important variables but altered body dimensions could also influence other physical characteristics, such as muscular strength.

These aspects are particularly important in regards to psychosocial issues, as under development of the physical characteristics can cause a psychosocial burden and affect wellbeing [28, 29].

General intellectual capacity

When it comes to people with cleft lip and palate and their cognitive abilities there are findings that has established that this group has cognitive deficits in comparison with the general population [30-34]. The reason for these deficits has either been attributed to the type of cleft [31, 35, 36] and individuals with cleft palate only manifest the most serious cognitive deficits. Studies that has examined the severity of the cleft [30, 37, 38] indicates that the severity is related to the extent of cognitive or developmental problems. These have been examined predominately in infants and children of school-age and indicate that there are cognitive deficiencies for the children with craniofacial anomalies, while there have been fewer studies on adolescents and adults but with the same indications [39].

Academic achievements

Education is one of the principal stepping-stones in the life of an individual and it has a direct influence on which pathways in life that will be accessible. Individuals that do not succeed in education, especially at the compulsory or upper secondary level, encounter more limited and arduous pathways of life, especially in relation to obtaining employment and future earnings power [40, 41], but aspects such as enhancement of well-being and quality-of-life can also be affected negatively [42]. Research has actually indicated that some individuals with cleft do experience cognitive dysfunction, learning difficulties, and lower school achievements in comparison to control groups [30, 33, 43, 44]. Since education is such an important component in life, it becomes interesting to more clearly analyze how adolescents with cleft manage their educational endeavors.

AIMS OF THE STUDY

The principal focus of this thesis was to examine if there are other variables that could theoretically contribute to the risk of psychosocial malady besides the cleft itself. Even though the first study focused upon psychological aspects in direct relation to the cleft, the study itself was important in the development of the idea that other variables could be a contributing factor that should be factored in when conducting research in this field.

The specific aims of the studies were:

- To investigate if adolescents with cleft lip and palate have an altered self-concept, and to assess their degree of introversion.
- To ascertain possible differences in weight, stature, body mass index (BMI), and muscular strength of young with cleft lip and palate in comparison to a control group.
- To establish if there is a difference in general intellectual capacity of young with cleft lip and palate in comparison to a control group.
- To determine if there is a difference in academic achievement of adolescents with cleft lip and palate in comparison with the general population.

MATERIALS

Article I

The purpose of this study was to investigate if cleft lip and/or palate adolescents have an altered self-concept, and to assess their degree of introversion, in comparison with a control group. The cleft lip and/or palate group consisted of 55 adolescents (17 to 20 years) while the control group consisted of 31 adolescents (16 to 19 years).

Article II and article III

The objective of article II was to report the weight, stature, body mass index (BMI), and muscular strength of men around 17 - 19 years old with cleft lip and palate, while article reports on their general intellectual capacity. Data were obtained from the Swedish National Service Enrolment Register for the years 1991-97, and concerned 335 men with cleft lip with/without palate and 88 with cleft palate, who were compared with a control group of 272 879 men.

Article IV

This paper reports on academic achievement at the time of leaving compulsory education (usually at 16 years old) for individuals with palate, individuals with cleft lip and individuals with cleft lip and palate. For this study, data were obtained from the Swedish Medical Birth Register for the years 1973-86 that was linked to the Swedish School – Grade Register, concerning 511 individuals with palate, 651 individuals with cleft lip and 830 individuals with cleft lip and palate, who were compared with a control group consisting of 1 249 404 individuals.

METHODS

Procedures

In article I, data were retrieved from the patient registers of the Department of Plastic and Reconstructive Surgery at Malmö University Hospital. The patients all had a cleft lip, a cleft lip and palate, or a cleft palate, and their age ranged from 17 to 20 years. Patients with any additional significant disability were excluded. One-hundred-and-twenty-five patients qualified for participation in the study. By mail they received the questionnaires, instructions for the tests, and a letter signed by members of the cleft team. The letter explained the purpose of the study, that participation was voluntary and anonymous if desired, and that refusal would not influence treatment in any way. Enclosed was also a return envelope with prepaid postage. Fifty-five patients, 27 males and 28 females with a mean age of 18.5 years, responded by completing the tests in their home environment, and mailing them back to us. Twenty-one of them had had a cleft lip and palate, 16 an isolated cleft lip, and 16 an isolated cleft palate. In two cases of anonymous reply, the cleft type was not clearly given, and consequently their scores were only used for the entire cleft lip and palate group.

The control group consisted of 31 adolescents, 11 males and 20 females, attending a local high school. Their ages ranged from 16 to 19 years with a mean of 17.5. The controls answered the questionnaires in a classroom setting during a psychology lesson. They got both written and verbal instructions by the teacher. Test-forms were collected by the researchers.

For article II and III, the medical data regarding physical and intellectual capacity were obtained from the National Service Enrolment Register for the years 1991-97 administered by the National Service Administration. Permission to use the register was obtained from The National Board of Health and Welfare [45] and all data about patients were unidentifiable. Military enrolment is mandatory for all Swedish men at the age of 17 – 19 years. Those not required to participate in military enrolment have either a severe handicap or a chronic disease that is documented medically and judged to permit exemption from any kind of military service. Men that have emigrated are also excluded. From this register, 335 people with repaired cleft lip with/without palate and 88 with repaired palate were identified. They were compared with a control group comprising 272 879 individuals. Neither in the cleft group nor in the control group were any men found to have additional handicap or disease (other congenital malformations or chromosomal abnormalities).

For article IV, the cases analyzed have been identified among adolescents born in 1973 – 1986 and registered in the Swedish Medical Birth Register (MBR) [45, 46]. The MBR contains data on pregnancy, delivery, and neonatal period, with links to information on deaths and migration. Additional cases and specification of cleft type has been achieved by utilizing The Register of Congenital Malformations. MBR has been linked to the Swedish School – Grade Register, which contains the final grades of all adolescents graduating from compulsory school, which is usually the year they turn 16 years old [47]. The cleft cases could be identified by the following characteristics: date of birth, hospital, birth time, and birth weight. The School – Grade Register is then linked to the cleft file without any use of personal identification and the analysis is conducted on an unidentifiable material. As in article II and article III, permission was obtained from the National Board of Health and Welfare to use the registers.

The analysis focused only on isolated clefts, no cases with associated malformations or syndromes were included. Among 2087 adolescents identified with an isolated cleft during these years, information from the School - Grade Register was obtained for 1992 (95.4%). Some cases could not be matched with the School – Grade Register due to errors of identification, emigration, or death before the age of 16.

Among the cleft adolescents, 511 had cleft palate, 651 cleft lip, and 830 cleft lip and palate. They were compared with the population consisting of 1 249 404 adolescents registered in MBR during the study period.

Questionnaires (Article I)

The Tennessee Self-Concept Scale

The Tennessee Self-Concept Scale (TSCS) was used to assess self-concept [48]. The Swedish version was obtained at the Department of Psychology, University of Lund, Lund, Sweden. The test consists of 10 self-descriptive dimensions, and of them nine different dimensions of TSCS was used. One is the total positive score reflecting the overall level of self-esteem of the subject. Three dimensions describe basic identity, perceived self-satisfaction, and perception of behavior. Other dimensions deal with physical self: health and appearance, sexuality and skills; and ethical self: moral worth, relationship to God, religious confidence, and feelings of being good or bad. Personal self deals with personal worth, adequacy as a person, relationship with others, whereas family self involves feelings of worth, adequacy, and value as a family member. Social self, finally, measures sense of worth, adequacy, and value in a wider social context. The standard norm of TSCS is based on 626 subjects representing an even distribution over age, socioeconomic factors, educational levels, and race [48]. The reliability for

the total positive score is .92. The TSCS is widely used and considered accurate, although qualities related to extroversion and anxiety may influence the outcome [49].

Eysenck Personality Questionnaire Inventory

Eysenck Personality Questionnaire Inventory (EPQ-I) was used to measure the degree of introversion [50]. The Swedish version was obtained at the Department of Psychology, University of Lund, Lund, Sweden. The test consists of all together 114 items, but only three scales are used for introversion-extroversion. Impulsiveness, sociability, and "unclassified" were combined to a total extroversion score. Consequently, a low score suggests introversion, and a high extroversion. The norm is based on 540 English males and 590 females aged 16 to 19 years (8). The internal consistency reliability is .85 for males and .84 for females. The test-retest reliability is .90 and .87, respectively [50]. In EPQ-I, cross-cultural studies have been yielded cognate results in several countries worldwide [51], which is an indication that the introversion/extraversion dimension is biological related [52].

Physical characteristics (Article II)

The following physical characteristics were analyzed: body weight (kg), height (cm), body mass index (BMI): $\text{weight (kg)/height (m)}^2$. BMI is an index of bodily composition used as a guide to assess if a person is overweight, normal, or underweight: $> 27 = \text{obese}$; $27 \text{ to } 25 = \text{overweight}$; $24 \text{ to } 20 = \text{normal}$; $19 \text{ to } 18 = \text{thin}$; and $< 18 = \text{undernourished}$. The last physical aspect analysed was muscular power (N), which was analysed by scoring muscular strength from tests on three muscular groups: hand grip, knee extension, and elbow flexion.

Swedish Enlistment Battery (SEB) (Article III)

The SEB is comprised of three primary factors; the induction, verbal comprehension and spatial visualisation. A fourth factor was added to measure technical comprehension, which makes the SEB a four – test – battery [53] where each factor is comprised of 40 items [54]. The four factors are summarized into a general score that indicates a general intellectual capacity [53]. A standard normal distribution is utilized; a scale from 1-9; where 9 indicates the best possible result, 6 represent the median (and mode) value in the population.

Academic achievement (Article IV)

Compulsory school is mandatory in Sweden for 9 years with an autumn and spring term each school year. The study analyzed the academic outcome from 1988 – 2003. Within this period the Swedish grading system changed. In the period 1988 – 1997, a relative grading system was applied. The system was based upon the mathematical theory of normal distribution and its main purpose was to rank the students. In this system, a 5 level grade scale was used, 1 was the lowest and 5 was the highest. The grades were distributed so that 7% of the students received grade 1; 24% grade 2; 38% grade 3; 24% grade 4; 7% grade 5 [55].

In 1998, a knowledge-related system was implemented, which means that the grades are based upon the basis of the target met by the students in accordance to the criteria of the syllabus. In this system, the student can achieve the following grades; Pass, Pass with distinction and Pass with excellence; furthermore, if the student does not meet the requirements no grade is issued in that subject [56], defined as Not Passed in this study.

The following aspects were analyzed:

1. Not receiving leaving certificate
2. Odds of receiving lowest grade and reduced odds in receiving high grade in the following subjects;
 - Mathematics
 - English
 - Swedish
 - Physical education
3. Grade Point Average (based only upon the numerical grades 1988 – 1997)

Statistical analysis

In article I, the differences between groups were analysed using the *t*-test and one-way analysis of variance (ANOVA). Self-concept and introversion were compared using linear correlation. In simple terms, the *t*-test compares the actual difference between two means in relation to the variation in the data (expressed as the standard deviation of the difference between the means), while the one-way analysis of variance is used when there are two or more independent groups to be analyzed. The linear correlation (Pearson's correlation coefficient [*r*]) was applied to measure the relation between to continuous variables in two methods [57].

In article II, III and IV, variables were divided in intervals in each of which comparisons were made between groups by means of Mantel-Haenszel procedure [58]. Risks are presented as odds ratios (OR) with 95% confidence intervals (95% CI) estimated by a test based method [59]. A procedure *t*-test was also used in article IV for each stratum (see below) and then a final *t*-test was performed.

In article II and III adjustments were made for year of birth, while in article IV the following adjustments were made; child's birth year, the mother age at child's birth, the mother's parity and the mother's educational level on a 7-grade scale.

RESULTS

Self-Concept and Introversion

In this study, TSCS indicated the individuals with cleft did not experience low self-concept in comparison with the control group, in actuality the overall score indicated a significant higher self-esteem 356.60 ± 30.22 ($t_{(80)} = 2.41$; $p < .05$) as well in sub-scale; identity, behaviour and family self. When comparing the individuals with cleft lip and palate, cleft lip or cleft palate, they did not differ from one another ($F_{(2,47)} = 1.79$; $p > .05$).

EPQ-I scores were 13.29 ± 4.90 in the control group, and 13.62 ± 4.04 in the group with cleft ($t_{(84)} = -0.33$; $p > .05$). Both scores are compatible with the introversion-extroversion normative group; 14.46 for males, and 13.31 for females [50]. Scores of subgroups; CLP, CL and CP did not differ from one another ($F_{(2,50)} = 0.79$; $p > .05$).

Scores in TSCS and EPQ-I showed no correlation ($r = 0.030$), neither in any of the subgroups.

Physical Characteristics

The findings are described in the following sections; body weight, height, body mass index and muscular strength for the groups with cleft lip with/without palate and cleft palate in relation to the control group.

Body weight

The OR for weighing less than 60 kg for the group with cleft lip with/without palate ($n = 321$) was significantly increased: 1.74 (95% CI 1.28 to 2.37) $p = 0.0002$, $\chi^2 = 13.55$. The same phenomenon was observed in the group with cleft palate ($n = 80$), the corresponding OR was 2.13 (95% CI 1.18 to 3.83) $p = 0.007$, $\chi^2 = 7.29$.

Height

The group with cleft lip with/without palate ($n = 321$) showed no significant deviation in height, the OR for being less than 170 cm was 1.21 (95% CI 0.77 to 1.89) $p = 0.22$, $\chi^2 = 0.80$. In the group with cleft palate ($n = 80$) there was a significant difference compared with the control group, the corresponding OR being 2.25 (95% CI 1.09 to 4.50) $p = 0.014$, $\chi^2 = 6.05$.

Body Mass Index

There was a significant difference between the group with cleft lip with/without palate (n=321) and the controls. The OR for having a BMI <19 was 1.55 (95% CI 1.12 to 2.12) p=0,005, $\chi^2=7,90$, in the group with cleft palate (n=80) there was no significant difference, the OR being 1.71 (95% CI 0.90 to 3.18) p=0,07, $\chi^2=3,19$, compared with the control group.

Muscular strength

Muscular strength was categorised in five levels and there was no significant reduction in muscular strength in the group with cleft lip with/without palate (n=316), the OR for muscular strength below the 1959 N being 1.22 (95% CI 0.97 to 1.53) p=0,08, $\chi^2=3,07$. In the group with cleft palate (n=88) there was an obvious reduction in muscular strength. The OR for muscular strength below the 1959 N was 1.59 (95% CI 1.00 to 2.55) p=0,04, $\chi^2=4,25$.

General Intellectual Capacity

The group with cleft lip with/without palate (n=307) showed no significant deviation in general intellectual capacity (GIC) in comparison to the control group. The OR for having a GIC score below 4 was 1.11 (95% CI 0.84-1.45) p=0,45, $\chi^2=0,57$. In the group with cleft palate (n=81) there was a significant reduced general intellectual capacity compared with the control group, the corresponding OR was 1.98 (95% CI 1.22-3.19) p=0,003, $\chi^2=8,85$.

Academic Achievement

The adolescents with cleft lip experience the least negative consequence, they have a significantly lower grade point average (Table I) and significantly higher odds of receiving lowest grade in math based upon the relative system OR 1.65 (95% CI 1.02-2.68), while they have significantly reduced odds of receiving the highest grades in Swedish based upon the knowledge based system OR 0.72 (95% CI 0.53-0.96). Nevertheless, they finish compulsory school and receive their leaving certificates to the same extent as the general population (Figure I).

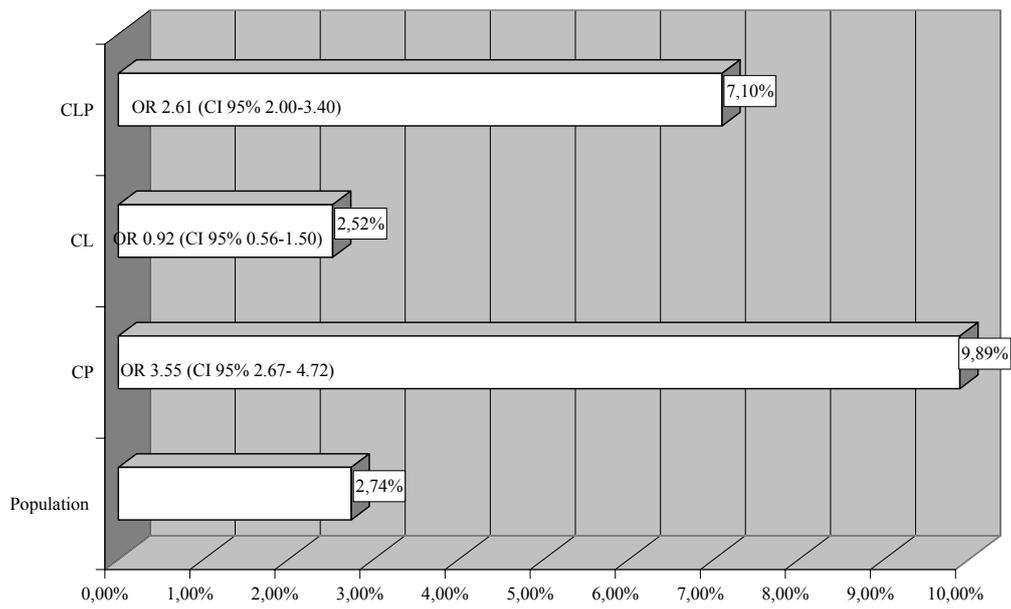
For the adolescents with cleft lip and palate, they experienced reduced odds of receiving the highest grades in English OR 0.80 (95% CI 0.65-0.99) and physical

education OR 0.78 (95% CI 0.64-0.96) as well as a significantly lower grade point average (see table I). Moreover, this group had a significantly higher rate of not receiving their leaving certificate (Figure I).

The adolescents with cleft palate experience the most negative impact; in the relative based system they had significantly high odds of receiving the lowest grade in mathematics OR 2.13 (95% CI 1.23-3.70), English OR 3.19 (95% CI 1.97-5.16) and Swedish OR 2.31(95% CI 1.29-4.16), while they had significantly reduced odds in receiving a high grade in Swedish (grade 5) OR 0.46 (95% CI 0.24-0.89) and physical education (grade 4) OR 0.74 (95% CI 0.55-0.99). In the knowledge based system they had significantly reduced odds in receiving high grades in mathematics OR 0.67 (95% CI 0.47-0.93) and physical education OR 0.63 (95% CI 0.46-0.86). They had a significantly lower grade point average (see table I) and significantly high rate of not receiving their leaving certificate in comparison to the population (see figure I).

| Table I. Grade point average (based only upon the numerical grades 1988 – 1997) | | | | | | |
|---|-----|-----------|------------|------------|--------|-------|
| Cleft type | n | Cleft | Population | Mean t | z-test | P |
| CP | 274 | 3.06±0.04 | 3.24±0.001 | -0.25±0.09 | -2.87 | 0.002 |
| CL | 428 | 3.12±0.03 | 3.25±0.001 | -0.19±0.07 | -2.78 | 0.003 |
| CLP | 471 | 3.12±0.03 | 3.24±0.001 | -0.17±0.06 | -2.71 | 0.003 |

Values after adjustment for year of birth, maternal age, parity and maternal education



*Figure I. Percentage not receiving leaving certificate
 Values after adjustment for year of birth, maternal age, parity and maternal education.*

GENERAL DISCUSSION

Methodological considerations

In study no. I on self-concept and introversion, fifty-five out of 125 individuals of the CLP group responded to our inquiry. This response rate of 44 percent is surprisingly good considering the sensitive subject of psychological testing [60]. An average return rate in initial surveys actually is 20 to 30 percent [61]. Considering the study design and the fact that primary surgery was done roughly 15 years earlier, it was decided not to contact the subjects a second time. The patients filled out the forms on their own in their various home environments. Obviously this did not involve any major problems, as only three forms were incomplete. Whenever possible from a practical point of view, like for our control group, the structured classroom setting is preferred where also supplementary oral instructions can be given. The cleft lip and palate group and the control group were compatible in regard to age and gender.

The strength of studies II and III is that the observations are based on a very large sample using the Swedish National Service Enrolment Register. However, a shortcoming is that only men were included. It is important to explore further whether physical characteristics are also altered in young adult women with cleft lip and palate and cleft palate. The same is true for the general intelligence capacity since it is established that girls with cleft palate, for instance, have learning difficulties [43]. These studies would require a different design as there is no corresponding database available for women.

Also study no. IV utilizes a large sample, in this case derived from the Swedish Medical Birth Register linked to the Swedish School-Grade Register. Furthermore, for the time period analyzed it was possible to include 95.4% of all children with clefts and compare them with all students during that period.

The registers used in study IV contained 229 cases of associated malformations or syndromes after exclusion of deceased or migrated adolescents. These individuals with associated malformations or syndromes were excluded. However, we investigated the percentage distributions between the different cleft types of associated malformations or syndromes, and they were; CP: 13%, CL: 7% and CLP: 11%. This is in the low end compared with a report by Sárközi et al based on a summary of other studies on the prevalence of associated anomalies; CP: 13-50%, CL: 7-13% and CLP 2-26% [62]. The reason for these variations can be linked to methodological issues, which is further outlined by Wyszynski et al [63].

Anyhow, central registers not primarily aimed at research of this kind may not be reliable to a hundred percent. This is of particular significance for individuals with

various types of clefts as they represent heterogeneous groups. An associated malformation or even a syndrome might have been overlooked in the screening procedures for the Swedish National Service Enrolment Register and the Swedish Medical Birth Register.

Self-concept and Introversion (Article I)

Findings in study no. I indicate that adolescents with cleft lip and palate have a good self-concept which is congruent with other comparable research [10, 11, 14, 64]. Compared with the control group, the cleft lip and palate group had in fact a higher mean value on all the TSCS scales, and there was a significant difference in three subscales and the overall self-concept score. The distribution of the scores was similar in the two groups, which go against that some cleft lip and palate individuals would be at particular risk for psychosocial problems. Such a risk has been postulated in younger adolescents with cleft lip and palate [11].

Neither were there signs of introversion in the cleft lip and palate group. This finding differs partly from previous reports demonstrating social introversion sometimes accompanied by poor self-concept and anxiety [13, 14]. The discrepancy can at least to some degree be explained by differences in methodology and subjects. The EPQ-I is based on the theory that biological factors are involved in personality dimensions like introversion/extraversion [52] whereas Richman's study for example [14] was based on the subjective view about the facial appearance rather than the primary cleft morphology. Pertschuk and Whitaker [13] studied children with craniofacial deformities such as craniofacial dysostosis, hemifacial microsomia, orbital hypertelorism, and mandibulofacial dysostosis, rather than adolescents with cleft lip and palate.

It is peculiar, though, that adolescents with cleft lip and palate manage to maintain their healthy self-concept in a society where beautiful is considered equal to good [65] and attractive people thought to possess other positive qualities as well [66]. The fact that a face with a repaired cleft is judged significantly less friendly and attractive and that the person is viewed less popular and unlikely to be picked as a friend [12] makes the situation still harder. However, individuals with cleft lip and palate may actively use their stigmatised status to maintain a healthy self-concept [67]. In other words, the stigma becomes a protective shield against negative feedback for the self-concept [68, 69]. Individuals with cleft lip and palate may also, or alternatively, use benign defense mechanisms to protect their self-concept from negative feedback, stigmatism, and social rejection [10, 13].

No data were obtained aimed at elucidating for instance socioeconomic status, general competence, school achievement, and confidence with the reconstructive result. This may be considered a shortcoming since it does not take into account other variables or determinants that has a crucial influence on the psychosocial wellbeing [7]. The following studies in this thesis therefore focus upon identifying other potential variables that could have an impact on the psychological or quality-of-life measure.

Physical Characteristics and Physical Education (Article II and IV)

In regards to physical characteristics, our results indicated that those in the cleft lip and palate group were significantly more likely to have a BMI less than 19, because they had a tendency to be thinner than normal, while the normal BMI in the cleft palate group was the result of both shorter stature and lighter weight. We suggest therefore, that there is a long term consequence of having a cleft and it is not apparent that those young men will reach the general norm for body dimensions. Interestingly, previous reports have indicated that, when growth is less among people with clefts, this might be more apparent in women than men [25, 26].

We have shown that stature and weight are affected by the presence of a cleft and that the effects differ between subjects with cleft of the primary palate and those with cleft of the secondary palate. This strongly supports other studies that have shown that the type of cleft does affect weight and stature [22, 23]. However, there are other studies showing that neither cleft type nor its severity has any bearing on these characteristics [19-21]. These results can be attributed to differences in classification and sample size. In the study by Bowers et al. [22], for example, nine categories of clefts were used, whereas in the present study we used only two groups. It would have been interesting to distinguish between cleft lip and cleft lip and palate in the cleft lip and palate group, but this was not feasible because of the way the registers were laid out.

There was no significant difference between the muscular strength of the cleft lip and palate group and the control group. In the cleft palate group, however, there was a significant reduction. This may be attributable to the fact that subjects in the cleft palate group were lighter and shorter because their body dimensions were less, with less muscular mass, which has been shown to be associated with reduced strength [70]. These alterations in body dimensions and muscle strength may explain the findings in study IV where adolescents with cleft lip and palate and cleft palate at school experienced reduced odds of receiving the highest grades in physical performance of their education.

Our findings indicate that cleft lip and palate and cleft palate have an effect on the physical characteristics of these young adult men. It would be interesting to examine how such physical consequences affect them psychosocially. Such an effect would be particularly relevant in ‘western’ society where physical beauty in accordance with certain cultural gender norms evoke a likable stereotype [71, 72]. This perception might be hard for patients with cleft lip and palate and cleft palate to emulate for those with a body composition that differs from the average population, as shown at least in young men. Physical attributes are an essential part of body image. They affect emotions, thoughts, behaviour and relationships [73]. Patients have also to cope with their aesthetic and functional defects associated with clefts. Taken together, these factors could generate a psychosocial liability and so affect their wellbeing [28, 29]. Attention also to physical characteristics may therefore constitute an expanded base for clinicians in their complete care of patients with clefts.

General Intelligence Capacity (Article III)

Previous studies have indicated that cognitive deficits exist in persons with non-syndromic clefts of the lip and/or palate [30-34]. The reason for these deficiencies are either attributed to the type of cleft [31, 35, 36] or severity of cleft [30, 37, 38]. The result of this study showed that the group with cleft palate had a significant lower score on the GIC compared with the control group, whereas the group with cleft lip and palate displayed no significant difference.

For the group with cleft palate, our result is in agreement with the results of other studies which indicate the coexistence of cognitive deficits [30, 31, 35, 36] and it is established that this group encounters learning disabilities [36, 39, 43].

Regarding the group with cleft lip and palate, our result contrasts to the results of previous research, for instance by Nopoulus et al. [30]. However, this discrepancy could be attributed to methodological differences since in their study the group with cleft lip and palate consisted of individuals with both cleft lip and cleft palate, whilst in this study the group with cleft lip and palate consisted of both individuals with cleft lip only and cleft lip and palate, simply due to the fact that our registers were not able to distinguish these two groups from one another.

The outcome of our study strongly supports the hypothesis that there is deficits in general intellectual capacity for the group with cleft palate alone and it would be interesting to examine in future studies how people with these disabilities respond in educational settings.

This became the purpose of our 4th study.

Academic Achievement (Article IV)

The study clearly indicates that adolescents with cleft experience significant difficulties in their educational achievements in compulsory school in Sweden, which is congruent with the studies that display that cognitive dysfunction, learning difficulties and lower school achievements [30, 33, 43, 44]. Furthermore, the study also supports the notion that the type of cleft has an influence on the outcome [31, 35, 36].

Adolescents with cleft lip experienced the least negative consequences. They were less capable in mathematics (relative system) and Swedish (knowledge based system), and they had a significantly lower grade point average compared with the control group (relative system). Nevertheless, they finished compulsory school and received their leaving certificates to the same extent as the general population.

Consequences for adolescents with cleft lip and palate were slightly more serious. They were less capable in English and physical education, and they had a significantly lower grade point average (relative system). More importantly, however; they had a significantly higher rate of not receiving their leaving certificate, 7.1% compared with 2.7% in the general population.

Adolescents with cleft palate experienced the most negative impact. In the relative based system, they were less capable in mathematics, English and Swedish as well as in physical education. This was also true for mathematics and physical education in the knowledge based system. Furthermore, they had a significantly lower grade point average (relative system), and a significantly higher rate of not receiving their leaving certificate, 9.9%. In study III we noted lower general intellectual capacity among men with cleft palate alone at the age of 17 – 19 years. The findings in this study indicate that declined intellectual capacity in this group is seen already at the age of 15 – 16 years.

After compulsory school, the student applies to upper secondary programs. Their achievements, either quantified as a grade point average in the relative system, or a sum of the abilities in the knowledge based system, determine which educational program they can enter. If they have not received their leaving certificate or a passing grade in mathematics, English and Swedish they can only enter an individual program, which is to prepare the pupil for studies on a national or specially-designed program. The present study clearly suggests that adolescents with cleft can experience a more difficult transition when continuing their education, and that a number of these adolescents are at a disadvantage in comparison with the general population. Consequently, it seems important to be aware of these problems so that additional

resources can be allocated in compulsory school whenever needed in order to guarantee the same educational achievement for adolescents with cleft as their peers. This reasoning seems even more relevant since some studies indicate that adults with cleft not only experience lower educational level [74, 75], but also a higher rate of unemployment [74] and a negative impact on their future earnings [74-76].

It is paramount for any young individual to attain educational achievement to avoid being in risk for marginalization in the society [77]. Consequently, it would be interesting to further examine academic achievement in upper secondary schools and unemployment rates for individuals with cleft since these factors have an influential impact on the quality of life.

CONCLUSION

- Adolescents with cleft seem to have a normal to high self-concept without an accentuated degree of introversion (I).
- A cleft influences body dimensions, particularly in the cleft palate group where weight, stature as well as muscle strength were reduced (II). Adolescents with cleft palate were also found to be less capable in their physical education at school (IV).
- Regarding the measured general intellectual capacity, adolescents with cleft lip with/without palate compared to the controls, whereas a significantly reduced score was noted in the cleft palate group (III).
- Academic achievement of adolescents with cleft is significantly reduced, particularly in the group with cleft palate (IV).

FINAL THOUGHTS AND REMARKS

The goal of modern cleft lip and palate treatment is to restore normal function and appearance. Important working tools in this endeavour are meticulous surgery regarding technique and timing, orthodontic treatment, speech therapy and care of hearing. Despite these attempts, complete normality is seldom achieved, but the individuals with clefts are merely without exception confronted with a deficit of some sort and to some degree. That stated a considerable proportion cope well [78] and based on our broad experience of these patients during a long period of time we also believe that most of them manage to cope with their situation in a fruitful way resulting in good psychological health, good social adaptation, normal education and acceptable professional life, this notion that a substantial proportion cope well. However, on the group level they do not fully compare to the normal population as elucidated in the present studies. Altered body dimensions reducing physical capacity may, for example, also contribute to the feeling of being different and then the obviously noticed educational difficulties indicates another complex factor. Consequently, it is important to be aware of such factors so that supportive measures can be instituted at an early stage as a complement to the ordinary rehabilitation programme.

SUMMARY

It is postulated that individuals with cleft may be at risk to experience psychosocial problems based upon their congenital malformation. Since cleft is one of the more common congenital malformations it is of importance to address this postulation. A fundamental question arises, however – is it right to assume that an individual with cleft will have an increased risk of psychosocial problems solely based upon the cleft? Even though psychological aspects in direct relation to the cleft may be important, other variables could be contributing factors that should be factored in when conducting research in this field. The following studies were therefore carried out:

In study I, 55 adolescents (17 to 20 years old) with cleft lip and palate and 31 adolescents (16 to 19 years old) without cleft filled out two questionnaires: the Tennessee Self-Concept Scale to assess self-concept, and the Eysenck Personality Questionnaire Inventory (EPQ-I) to assess the degree of introversion. In studies II & III, data regarding physical characteristic (weight, stature, body mass index, muscle strength) and general intellectual capacity (induction, verbal comprehension, spatial visualisation, and technical comprehension) were obtained from the Swedish National Service Enrolment Register for the years 1991-97. Data concerned 335 men with cleft lip with or without cleft palate and 88 with cleft palate. They were compared with a control group of 272 879 men without cleft. In study IV, data regarding academic achievement (leaving certificate, mathematics, English, Swedish, physical education, grade point average) at the time of leaving compulsory education (usually at 16 years old) were obtained by linking the Swedish Medical Birth Register for the years 1973-86 with the Swedish School Grade Register. Data concerned 651 individuals with cleft lip, 830 with cleft lip and palate, and 511 with cleft palate. They were compared with a control group consisting of 1 249 404 individuals without cleft.

We found that adolescents with cleft seem to have a normal to high self-concept without an accentuated degree of introversion (I). A cleft influences body dimensions, particularly in the cleft palate group where weight, stature, and muscle strength (II) showed lowered values, this may be correlated to their reduced achievement in physical education at school (IV). The general intellectual capacity (III) as well as the academic achievement (IV) were both mostly adequate for adolescents with cleft lip with/without cleft palate, whereas adolescents with cleft palate performed somewhat worse on average.

The first study followed the notion that an individual with cleft will have an increased risk of psychosocial problems completely based upon the cleft *per se*, which is an arbitrary notion since it does not take into account other variables. The subsequent studies therefore focused upon identifying other potential variables that could have an impact on the psychological or quality-of-life measure. Our findings in

study II indicate that cleft lip and palate and cleft palate have an effect on the physical characteristics of young adult men and attention also to these alterations may therefore constitute an expanded base for clinicians in their complete care of patients with clefts. The outcome of our third study strongly supports the view that there are deficits in general intellectual capacities for the group with cleft palate. Therefore, it seemed interesting to examine how persons with cleft, especially cleft palate, perform in educational settings in regards to academic achievement. Our last study actually showed that adolescents with cleft palate experienced notable difficulties in their educational achievements. Consequently, it is important to be aware of these problems so that additional resources can be allocated in compulsory school whenever needed in order to guarantee the same educational achievement for adolescents with cleft as their peers.

SUMMARY IN SWEDISH – POPULÄRVETENSKAPLIG SAMMANFATTNING PÅ SVENSKA

LÄPP-KÄK-GOMSPALT I UNGDOMEN

Faktorer av möjlig betydelse ur psykosocial synpunkt

Det kan finnas en risk för att enskilda individer med läpp-käk-gomspalt (LKG) kan möta problem ur psykosocial synpunkt som en konsekvens av den medfödda avvikelserna. LKG är en av de vanligaste medfödda avvikelserna och sett ut den synpunkten synes det viktigt att närmre undersöka detta antagande. Detta leder till den viktiga frågan om det är LKG-spalten som sådan som är avgörande eller om det även finns andra faktorer i bilden som spelar roll. Även om psykologiska och funktionella aspekter som är direkt relaterade till spalten kan vara viktiga, så valde vi att inom föreliggande arbete också belysa andra faktorer som skulle kunna bidra till en försvårad anpassning ur psykosocial synpunkt för den enskilde. Följande studier genomfördes därför:

I studie I fyllde 55 ungdomar (17 till 20 år gamla) med LKG och 33 ungdomar (16 till 19 år gamla) utan LKG i två enkäter; Tennessee Self-Concept Scale som uppskattar självkänsla, och Eysenck Personality Questionnaire Inventory som uppskattar introversion. I studierna II&III analyserades oidentifierade data angående fysiska karaktärer (vikt, längd, body mass index, muskel styrka) samt generell intellektuell kapacitet från mönstringsregistret under åren 1991 – 1997. Materialet inkluderade 335 män med läppspalt med eller utan gomspalt och 88 med gomspalt. Dessa jämfördes med en kontroll grupp på 272 879 män utan spalt. I studie IV analyserades oidentifierade data angående utbildningsresultat (avgångsbetyg, matematik, Engelska, Svenska, idrott, medelbetyg) vid avgång från grundskolan. Födelseregistret (1973-86) länkades därvid med betygsregistret och 651 ungdomar med läppspalt, 830 med LKG-spalt samt 511 med gomspalt kunde jämföras med en kontroll grupp på 1249404 ungdomar utan spalt.

Allt som allt noterade vi att ungdomar med LKG-spalt har en normal till en hög självkänsla och ingen märkbar skillnad i introversion (I). LKG påverkar de fysiska karaktärerna, särskilt inom gomspaltgruppen där vikt, längd, och muskelstyrka hade lägre mätvärden (II) och den något lägre prestationen i grundskolans idrottsämne kan ha samband med detta (IV). Den generella intellektuella kapaciteten (III) liksom den akademiska förmågan (IV) var genomgående adekvat för ungdomar med läppspalt med eller utan gomspalt, medan ungdomarna med gomspalt som grupp presterade något sämre.

Den första studien byggde på idén att en individ med spalt kan möta problem ur psykosocial synpunkt baserat på spaltens funktionella och utseendemässiga

konsekvenser. De följande studierna tog fasta på idén att även andra faktorer kan finnas med i bilden och påverka individens psykosociala hälsa och livskvalitet. I vår andra studie påvisade vi att fysiska karaktärer kan vara annorlunda vid spalt, något som också det kan påverka livssituationen. Resultaten av de tredje och fjärde studierna stödjer främst tidigare observationer att ungdomar med gomspalt som grupp betraktad har vissa kognitiva svårigheter som påverkar den intellektuella kapaciteten och förmågan att hävda sig akademiskt.

Sammanfattningsvis förefaller det angeläget att se ungdomar med LKG-spalt i ett vidare perspektiv än vad vi tidigare gjort så att extra resurser i enskilda fall där behov finns kan allokeras till skolan på ett tidigt stadium medgivande lika förutsättningar för alla vad det gäller utbildning och yrkesutövning.

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