

# Scientific Section

## SPECIAL ARTICLE

# The Clinical Standards Advisory Group (CSAG) Cleft Lip and Palate Study

JONATHAN SANDY, PH.D., M.ORTH, F.D.S.R.C.S. (ENG.), F.D.S.R.C.S. (EDIN.)

ALISON WILLIAMS, M.SC., M.ORTH., F.D.S.R.C.S. (ENG.)

SUE MILDINHALL, L.R.C.S.L.T.,

Division of Child Dental Health, University of Bristol Dental School, Lower Maudlin Street, Bristol, U.K.

TERRIE MURPHY, L.R.C.S.L.T.

DAVID BEARN, M.SC., M.ORTH., F.D.S.R.C.P.S. (GLAS.)

BILL SHAW, PH.D., D.ORTH., F.D.S.R.C.S. (ENG.), D.D.O.R.C.P.S. (GLAS.)

Department of Orthodontics, University Dental Hospital Manchester, Higher Cambridge Street, Manchester, U.K.

DEBBIE SELL, PH.D., L.R.C.S.L.T.

Hospital for Sick Children, Great Ormond Street, London U.K.

BRENDAN DEVLIN, C.B.E., F.R.C.S.

Royal College of Surgeons of England, London U.K.

JOHN MURRAY, C.B.E., PH.D., F.D.S.R.C.S. (ENG.)

Department of Child Dental Health, Newcastle Dental School, Newcastle upon Tyne, U.K.

**Abstract:** *A national study of care and outcomes in children born with a unilateral cleft lip and palate (UCLP) was performed over a 15-month period. Two cohorts of children ('5-year-olds' and '12-year-olds') were examined. There were 57 active cleft teams in the U.K. with 105 consultant orthodontists involved in the care of these children. Only 36 teams could provide basic data such as patients names. Of the patients, 47-51 per cent had neonatal appliances. The dental arch relationships were measured with the Goslon Index and a Five-Year-Old Index, 37-39 per cent of both age groups were either 'poor' or 'very poor'. Seventy per cent of the 12-year-old patients had a Skeletal III relation and 42 per cent of bone grafts were seriously deficient or failed. Dental treatment for active caries was needed by 40 per cent of 5-year-olds and 20 per cent of 12-year-olds. In addition, the training of recently appointed consultant orthodontists involved in the care of these children was scrutinized. As a whole, the results were disappointing with standards of care not significantly raised in the last decade. Recommendations have been made to the Department of Health and the implications for the orthodontic profession are explored. Overall, it seems that fewer orthodontists will need to be involved in a centralized care model for these children.*

*Index words:* Delivery of Cleft Care, Outcome Measures, Unilateral Cleft Lip and Palate.

### Introduction

An informative editorial in May 1996 (Jones, 1996) outlined an ambitious study commissioned by the U.K. Health Ministers to advise on standards of clinical care for children with congenital cleft lip and or palate (CLP). The

project, supervised by the Clinical Standards Advisory Group (CSAG), officially commenced on the 1st March 1996 with funding for 15 months. The report was produced following consideration of the data produced by the research team, with supporting visits to a number of units by the main cleft lip and palate Committee. After consideration of all the data available, and following lengthy discussion the report was completed and delivered on time.

---

Correspondence: Dr J. R. Sandy.

The CSAG cleft lip and palate committee had representation from all of the main disciplines involved in cleft care, including speech and language therapy, plastic surgery, oral and maxillofacial surgery, paediatric dentistry, nursing, and orthodontics. In addition, the chief executive of the Cleft Lip and Palate Association (CLAPA) was invited to participate as a full committee member. The main CSAG Committee were in receipt of the final report at the end of May 1997. This main committee represents all disciplines in and allied to medicine and dentistry, including all Royal Colleges, the chief medical officers in the U.K. and the Department of Health. The chairman of this main CSAG Committee was from a non-medical profession. At a meeting of the main CSAG Committee in June 1997, the report was accepted in full at its 'first offering'. This paper is intended to outline the role of the orthodontist in the project, whilst presenting some relevant findings. The potential implications of the recommendations from the report are also discussed.

## Methods

### *Appointment of Research Team*

Funding from the Department of Health provided the resource to support a whole time equivalent research registrar, speech and language therapist, and project administrator. To facilitate the management of such a large project the U.K. was divided into North and South Regions with 'half time' appointments in each region.

### *Network of Regional Cleft Co-ordinators*

In order to assist the research team with data collection a system of 'Regional Cleft Co-ordinators' was established, incorporating the network of orthodontic co-ordinators which already existed. A representative from the plastic surgery, maxillofacial surgery, and speech and language therapy specialities was nominated by local clinicians in each region to act as a 'regional co-ordinator' for their speciality. A 'lead co-ordinator' was then selected from the co-ordinators within each region to liaise between the local cleft teams and the research team. The regional cleft co-ordinators were invited to identify all functioning cleft teams within their regions to the research team.

A 'Cleft Team Questionnaire' was developed and piloted by the research team. The questionnaire was constructed to obtain information on facilities offered by the cleft team, including the provision of any 'outreach' clinics. Details were requested of the members of the cleft team including their surgical speciality where appropriate. Cleft teams were also asked to provide lists of the referring maternity units and the Purchasers holding 'block contracts' for cleft care. Details on the use of patient databases were requested together with the number of new referrals to the cleft team during the calendar year of 1995.

Each cleft team was sent a copy of the questionnaire by post together with a prepaid reply envelope. A reminder was sent to cleft teams who failed to return the questionnaire within the period designated by the research team. Persistent non-respondents were contacted by telephone.

### *Selection of the sample*

Children born with a complete unilateral bony cleft of the lip and palate including a soft tissue band of less than 5 mm were chosen for inclusion in the study. This enabled an assessment of total cleft care to be undertaken and to allow comparison with other multicentre studies. Two age cohorts were chosen:

1. '5-year-olds'—defined as children born between 1.4.89 and 31.3.91.
2. '12-year-olds'—defined as children born between 1.4.82 and 31.3.84.

The '5-year-olds' were chosen to represent the first point at which the outcome of primary surgery could be assessed. The second age cohort was chosen to show the results of secondary alveolar bone grafting. Syndromic cases and those children with developmental delay sufficient to prevent co-operation with record collection by the research team were excluded.

### *Identification of the sample*

The leaders of cleft teams were invited to give details to the research team of children within the study cohort currently under their care. In addition, the regional plastic and maxillofacial surgery co-ordinators nominated junior staff to undertake a search of the operating books relevant to cleft surgeons operating during the periods 1.4.82–31.12.84 and 1.4.89–31.12.91. The original diagnosis was confirmed, where possible, from pre-lip repair photographs and/or study models. If there was a doubt as to the suitability of the subject then the final decision on inclusion was made by the research team. Each child identified in the study cohort was allocated a unique ID number and invited to attend at a data collection centre.

### *Process Audit*

In order to gather information on treatment received to date by each child in the study, data collection forms were developed by the research team for:

1. Surgical details—including primary surgery, revision surgery and alveolar bone grafting.
2. Audiology—details of ENT treatment.
3. Speech—speech and language therapy.
4. Dental health audit—details of dental and orthodontic treatment, including the provision of neonatal appliances.

### *Protocols for the collection of outcome data*

The outcome of cleft care was assessed in terms of dental arch and skeletal relationships, success of alveolar bone grafting, facial aesthetics, speech, hearing, dental and psychosocial status together with patient satisfaction. The following standardized records were collected using previously agreed protocols:

- (1) audiology and tympanometry;
- (2) speech recording;

- (3) photographs;
- (4) dental study casts;
- (5) oral health status, including the presence of fistulae;
- (6) parent and patient (12-year-olds only) satisfaction;
- (7) radiographs of alveolar bone graft (12-year-olds only);
- (8) lateral cephalogram.

#### *Organization of the data collection days*

The lead co-ordinators were asked to identify three or four 'data collection' centres in each region to which it would be convenient for patients to travel for record collection. A local co-ordinator was identified from each data collection centre with whom the research team could liaise regarding arrangements for the record collection day. Where possible, a member of the research team made a preliminary visit to the centre 2–3 weeks beforehand to view the facilities and discuss the arrangements for the data collection day. A letter of support for the study from CLAPA was also sent to parents. If the appointment was inconvenient an alternative appointment was offered where possible. The family's travel costs were met by the Department of Health.

#### *Data Analysis*

*Process data.* The information recorded on the data collection forms for any surgical, ENT, orthodontic, and speech and language therapy treatment received was coded by the research team, and entered into a database SPSS (SPSS Inc., Chicago) for analysis.

#### *Outcome data*

*Dental arch relationship.* The study models cast from the dental impressions were duplicated, together with any existing study models, in an agreed standardized form by one of two designated technicians. The models were divided into the two age cohorts and arranged randomly on a workbench. The dental arch relationship of the 5-year-old models was assessed using the 5-Year-Old Index (Atack *et al.*, 1997) and the 12-year-old models assessed with the well established GOSLON index (Mars *et al.*, 1987).

Both sets of models were assessed on two separate occasions by two orthodontically qualified examiners who had been calibrated in the use of both indices. The models were re-randomized prior to the second assessment. A consensus view was derived from a consultant orthodontist and the two observers in any cases where there were discrepancies. The intra- and inter-examiner agreement was calculated using the weighted kappa statistic.

*Skeletal relationship.* All the lateral cephalostat radiographs were traced by a single observer. The skeletal relationship was calculated using the method described by Mølsted *et al.* (1992). In order to assess intra-examiner reliability, 30 radiographs were retraced after a period of 4 weeks.

*Success of alveolar bone grafting.* Two experienced orthodontists assessed the radiographs collected for children

who had received alveolar bone grafts. The quality of the bone graft was assessed using a modification of the protocol developed by Bergland *et al.* (1986). Four categories of outcome and a fifth category 'unreadable' were rated by two experienced orthodontists. The inter-examiner agreement was calculated using the weighted kappa statistic. Where any disagreement occurred the film was re-read by both examiners and the category agreed by consensus.

*Psychosocial status and patient satisfaction.* The responses of parents and patients to established questionnaires (Thomas *et al.*, 1997; Turner *et al.*, 1997) were coded by the research team and the information entered into the database for analysis.

#### *Current training arrangements for consultant orthodontists*

The aim of this part of the study was to investigate how the training was arranged for consultant orthodontists. A list of 12 consultant orthodontists who had recently been appointed to cleft teams was assembled. Each orthodontist was sent a questionnaire asking for details of the clinical experience of cleft care gained during registrar/senior registrar training. A pre-paid reply envelope was supplied. Details were requested of cleft courses attended, and their participation in research and audit as trainees. The newly-appointed orthodontists were also asked whether they felt that their training could have been improved and to give suggestions for future training of orthodontists working with cleft teams. In addition, a list of senior consultant orthodontists was compiled from orthodontists working with cleft teams in district hospitals and those working in teaching hospitals. The senior orthodontists were invited to give their opinion as to how the training of cleft team orthodontists should be organized in the future.

## **Results**

### *Cleft teams*

A total of 59 cleft teams which had recently been involved in cleft care were identified to the research team by the regional co-ordinators. Each team was allocated a unique ID number and sent a copy of the Cleft Team Questionnaire. Two of the questionnaires were returned stating that cleft repairs were no longer being undertaken at these centres. Therefore, there were 57 cleft teams currently involved in cleft care. Four teams had started operating on primary clefts after 1991 and, thus, had no patients for inclusion in the study. The number of cleft teams in each Region varied from one in Northern Ireland, to 10 in the Northern and Yorkshire Region. Two entirely separate cleft teams were based at one hospital.

Forty-eight cleft teams completed and returned a Cleft Team Questionnaire, a response rate of 84 per cent. Each cleft team held, on average, one (range 0–4) outreach multidisciplinary clinic in addition to clinics held at the cleft team base. Cleft teams were contracted to provide services for a mean of 4.2 purchasers (range 1–16) per team. Each team received referrals of babies born with clefts from an average of 5.0 maternity units (range 1–15). A total of 75 cleft surgeons were identified from the

responses to the questionnaire, supplemented by local knowledge. These included 56 plastic surgeons, 15 oral and maxillofacial surgeons, three paediatric surgeons and one ENT surgeon. Seventy speech and language therapists, and 105 consultant orthodontists were involved in cleft care and took part in multidisciplinary cleft clinics.

### Sample

A total of 647 children, 326 5-year-olds and 321 12-year-olds, were identified to the research team. These children were under the care of 50 of the 57 current cleft teams.

'Surgical Notes' forms were completed for 91 per cent of 5-year-olds and 86 per cent of 12-year-olds. In some cases details of the primary surgery undertaken for children were missing. This was either because the child had moved to another cleft team or the cleft team itself had moved to another hospital site. No details of alveolar bone grafting could be collected for 36 of the patients identified from the older age cohort. Although one would expect all children born with a unilateral cleft to have undergone secondary alveolar bone grafting by the age of 11, there was no record of this for 15 per cent of 12-year-olds.

Approximately half of the 5-year-olds and the same proportion of 12-year-olds had a neonatal appliance fitted as a baby. The majority of 12-year-olds had received orthodontic treatment. Most of the children were currently enrolled with a dentist (Table 1).

### Data collection

A total of 601 children were invited to attend data collection days: 239 5-year-olds and 218 12-year-olds attended. This comprised 76 per cent of those invited. The non-attenders fell into three categories: 37 refused, 43 cancelled, and 64 accepted an appointment, but failed to attend.

### Outcomes

**Dental arch relationship.** Study models were collected for 223 5-year-olds and 229 12-year-olds, including 11 sets of study models of 12-year-olds who were unable to attend a data collection day. The study models collected for the younger age cohort were ranked using the 5-Year-Old Index and the 12-year-old models were ranked using the GOSLON index. The kappa statistic for inter-examiner agreement (1.0 = perfect agreement) was calculated as 0.94 for the 5-year-old models and 0.92 for the 12-year-old models. Figure 1 shows that 37 per cent of 5-year-old models and 39 per cent of 12-year-old models were rated as 'poor' or 'very poor'.

Parents of both age groups were moderately satisfied with the appearance of their child's teeth as were the 12-year-old patients (Fig. 2).

### Skeletal relationship

Lateral cephalometric radiographs were collected for 215 12-year-olds. Table 2 shows the skeletal relationship as determined from angle ANB of 215 12-year-olds for whom lateral cephalograms were available. Seventy per cent had a Skeletal III relationship. The mean ANB for the whole sample was  $-0.4^\circ$ . Parents were generally satisfied with the appearance of their child's profile (data not shown).

### Quality of alveolar bone graft

The quality of bone grafts was assessed for 183 children who had undergone this procedure and had anterior occlusal radiographs available. A recent radiograph was also provided for one child who was unable to attend for data collection. Twenty-six radiographs were considered to be unreadable due to poor film quality or tooth position. Thus, radiographs were available for 157 children who had undergone alveolar bone grafting. Only 58 per cent of bone grafts were successful (Table 3).

TABLE 1 Details of dental and orthodontic treatment received by 5- and 12-year-old children born with UCLP attending data collection days

	5-year-olds <i>n</i> = 239		12-year-olds <i>n</i> = 218	
	Frequency	%	Frequency	%
Had a neonatal appliance	112	47	112	51
Enrolled with a dentist	226	95	209	96
Had orthodontic treatment	N/A	N/A	188	86

TABLE 2 Skeletal relationship in 12 year old children with UCLP

	Frequency	%	Range (degrees)	Mean
Skeletal I	36	17	+2→+4	2.9°
Skeletal II	29	13	+4→+8	5.6°
Skeletal III	150	70	-13→+2°	-2.4°

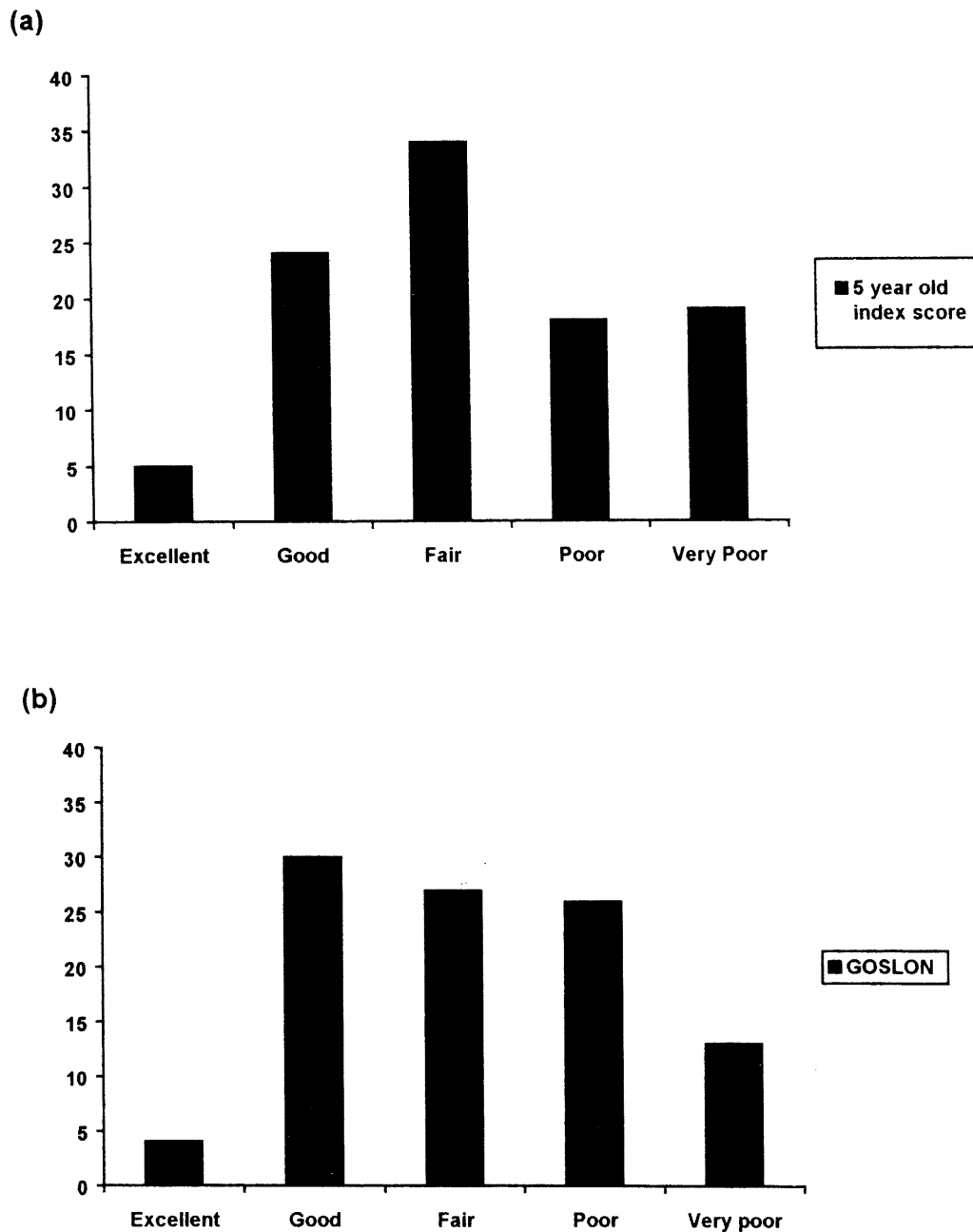


FIG. 1 (a) Dental arch relationships of 5-year-olds with UCLP. (b) Dental arch relationships of 12-year-olds with UCLP.

### Oral health

The research registrars examined the teeth of 239 5-year-olds and 218 12-year-olds. The mean dmf for 5-year-olds was 2.0 (range 0–18) and the mean DMF for 12-year-olds was 1.5 (range 0–8). Forty per cent of 5-year-olds were in need of dental treatment for caries, 20 per cent of 12-year-olds required treatment for dental decay. Thirty-nine per cent of 5-year-olds and 10 per cent of 12-year-olds were found to have a persistent oral fistula which was causing problems.

### Psychosocial status

Four-hundred-and-thirty parents completed the section of the questionnaire which was related to psychosocial issues. Nineteen per cent of parents of 5-year-olds and 28 per cent of parents of 12-year-olds felt that their child's self-confidence had been affected by the cleft. A third of the 12-year-olds attending the data collection day were being teased currently about the cleft and, of these, 25 per cent were worried by this teasing.

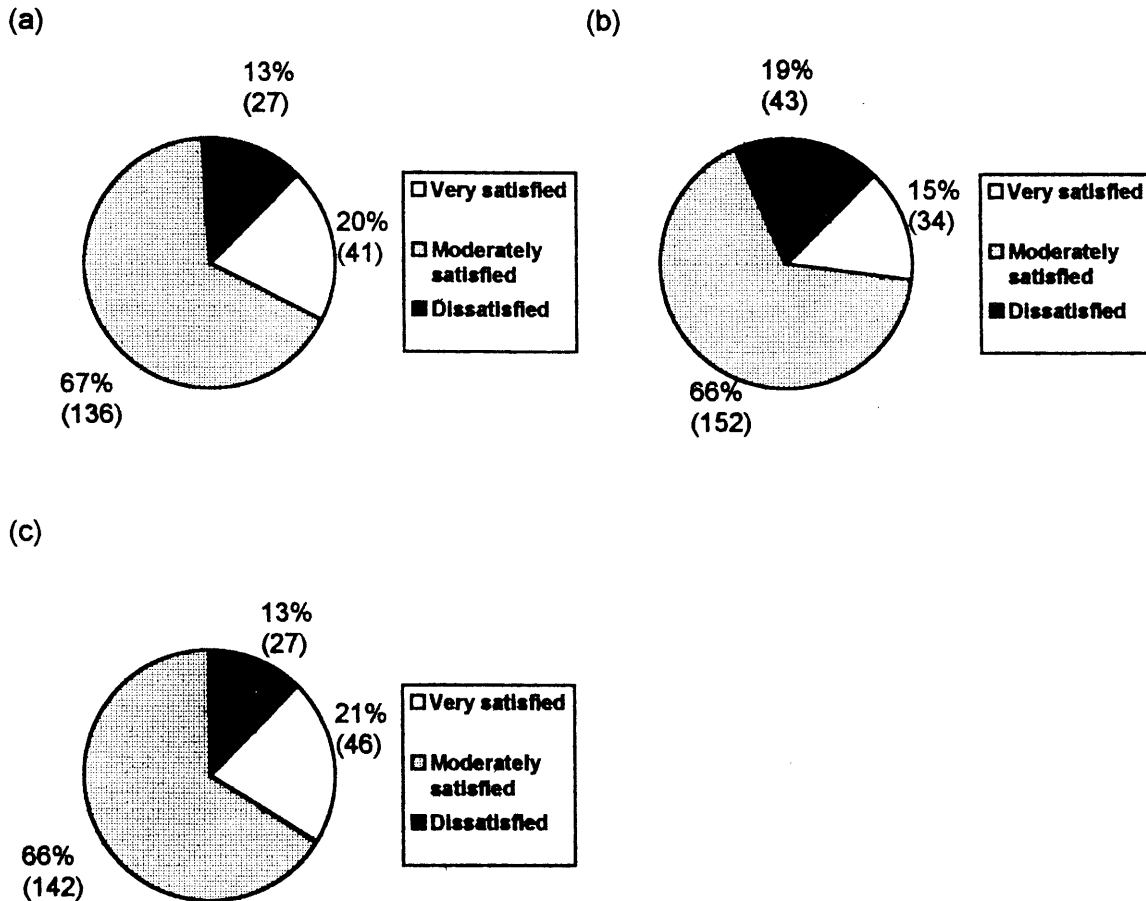


FIG. 2 Parent and patient satisfaction with appearance of teeth. (a) Parent satisfaction with the appearance of 5-year-olds teeth. (b) Parent satisfaction with the appearance of 12-year-olds teeth. (c) Patient satisfaction (12-year-olds) with appearance of teeth.

### Difficulties attending cleft clinics

Four-hundred-and-fifty-one parents completed the section on the questionnaire which referred to travel time to the cleft clinic and difficulties attending. Thirty per cent of parents lived more than 1 hour's travel time from home to the clinic. Difficulties in attending were reported by 36 per cent of parents. Not all of these difficulties were related to distance; other problems included taking time off work, arranging child care for siblings, and the child missing school.

### Parent satisfaction

As part of the questionnaire, parents were asked to rate their satisfaction with 'care and attention' received from the cleft teams, and the 'results and outcome' as two separate issues. Sixty-seven per cent of the 438 parents who completed this section of the questionnaire felt that they had received excellent care from the cleft team. Six per cent were dissatisfied. Fewer parents (56 per cent) felt that the 'treatment and outcome' of care had been excellent. Thirty-five per cent thought that the outcome of care was 'good', but 9 per cent were dissatisfied with the overall outcome (Fig. 3).

### Training of Orthodontists

Twelve consultant orthodontists who had been appointed to cleft teams within the past 5 years were surveyed. All 12 had M.Sc.'s and two had recently been awarded Ph.D.'s. Eleven responded. All of the recently appointed orthodontists had worked with cleft teams at both Senior Registrar and Registrar level. All had attended multidisciplinary cleft clinics as Senior Registrars.

The recently appointed orthodontists were also asked to give the number of cleft procedures they had undertaken at both Senior Registrar and Registrar level (Table 4).

All of the orthodontists surveyed had attended cleft courses as trainees, three of these had funded themselves. Eight of the recently appointed orthodontists had visited

TABLE 3 Outcome of alveolar bone grafting in 12-year-olds with UCLP (n = 157)

Outcome	Frequency	%
Successful	91	58
Seriously deficient	47	30
Failed	19	12

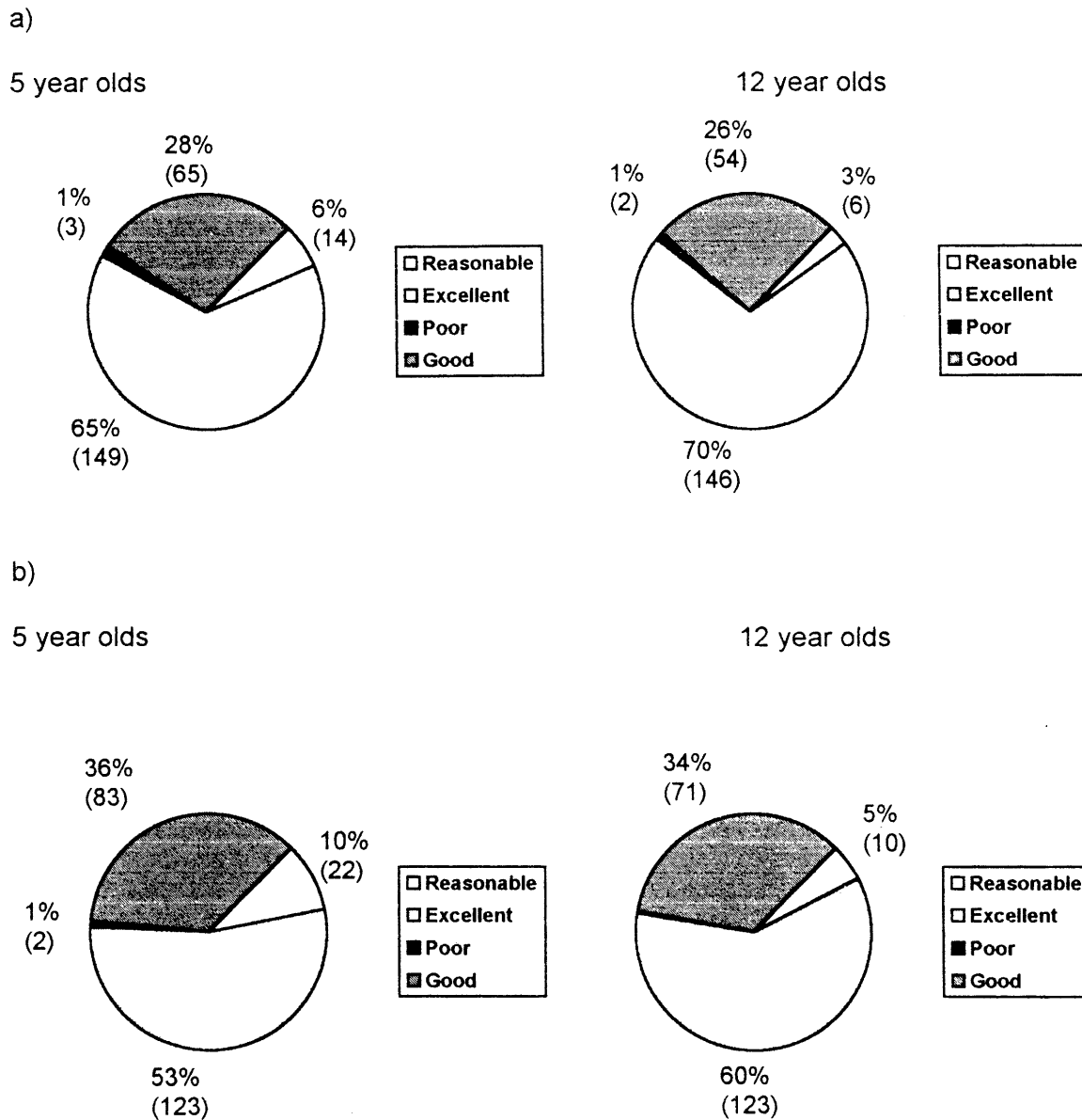


FIG. 3 Satisfaction of parents of 5- and 12-year-olds born with UCLP. (a) Parent satisfaction with care and attention, 5- and 12-year-olds. (b) Parent satisfaction with treatment and outcome, 5- and 12-year-olds.

centres overseas, three of these trips were self-funded. Only six respondents had been involved in cleft audit projects and less than half had published on cleft work. Sixty per cent of the newly appointed consultant orthodontists felt that their training could have been improved. More study days were requested together with more opportunity for personal treatment of a wide range of cases. When asked how future training of cleft team orthodontists should be organized, five of the respondents stated that clinical training should take place with large cleft teams. It was also felt important for trainees to visit other cleft centres in both the U.K. and abroad.

Five senior consultant orthodontists working in teaching hospitals and five orthodontists working in regional cleft

units gave their opinion on how future training for cleft team orthodontists should be organized. Half of the respondents felt that there should be identified training posts in high volume centres where trainees would be exposed to all aspects of cleft work. These posts should be linked to eventual consultant appointments in regional cleft centres where the consultant would be responsible for treatment planning on a 'hub and spoke' basis. Two respondents stated that cleft training for orthodontists should be formalized and one suggested that trainees should do an additional year of training. The majority of experienced cleft orthodontists thought that training should include research and audit together with close clinical supervision of cleft cases.

TABLE 4 Procedures undertaken by eleven newly-appointed consultant orthodontists during their training

Procedure	Registrar Undertook procedure (n = 11)		Mean number undertaken	Range	Senior Registrar Undertook procedure (n = 11)		Mean number undertaken	Range
	Frequency	%			Frequency	%		
Counselling	2	18	3	2-4	9	82	46.4	15-100
Neonatal impressions	5	46	3.7	2-5	10	91	24.3	5-100
Records for audit	3	27	4	3-5	10	91	45	20-100
Presurgical orthopaedics	1	9	1	1	7	64	16.6	5-50
Feeding plates	3	27	1.5	1-2	8	73	5.3	2-10
Pre- bone graft orthodontics	5	46	5	2-10	11	100	18.1	8-30
Definitive orthodontics	8	73	4.4	1-10	10	91	20.7	10-40
Preparation for orthognathic surgery	4	36	4.3	0-10	11	100	7.25	1-30

## Discussion

Following data analysis by the research team and their own visits to a sample of the cleft centres the CSAG Cleft Lip and Palate Committee made the following recommendations:

### *Recommendation to the U.K. Health Departments*

The U.K. Health Departments should ensure that the present arrangements involving 57 cleft units are changed so that expertise and resources are concentrated in 8-15 centres in the U.K. taking into account population needs and accessibility.

### *Recommendation to Purchasers/Commissioners*

Service specifications for cleft care should clearly indicate the range of expertise required in the team, the quality standards required in respect of process and clinical outcomes, and the information required for contract monitoring. Commissioners should purchase cleft care only from centres which fulfil these specifications.

### *Recommendations to Trusts/Provider Units*

Trusts currently providing cleft care should review their services and ensure that the full range of clinical skills needed are readily available.

Trusts should, in collaboration with purchasers and practitioners, develop plans for the concentration of cleft services.

### *Recommendations to Practitioners/Clinicians*

Clinicians should agree on a common database for all cleft patients, specifying information requirements and timing of collection, and ensure that all cleft patients are included. Information on all cleft patients should be made available for comparative audit studies.

### *Recommendations to Royal Colleges and Faculties*

Training programmes for all specialist cleft clinicians should be approved only in cleft centres where high volume of high quality clinical experience is available.

The relevant surgical specialities, plastic surgery, and oral and maxillofacial surgery must determine an agreed training pathway for the small number of surgical trainees required to specialise in cleft care.

### *Recommendation to the Office of National Statistics*

The completeness of recording of cleft births in the U.K. should be improved.

There is little doubt that, if implemented, these recommendations will have significant consequences, but the Consultant Orthodontists have already demonstrated great commitment to the care of children born with a cleft lip and palate. A previous survey yielded a 100 per cent response rate to a questionnaire sent to this group of consultants (Williams *et al.*, 1996). However, no previous National studies have examined outcomes on such a wide front and although there was no direct assessment of orthodontic results, there are areas of concern. The orthodontist often assumes a role in providing feeding advice, counselling, and ensures that adequate dental care is being provided. There is also a substantial role in record keeping and involvement in the stage of alveolar bone grafting. It is alarming then that 20-40 per cent of 12- and 5-year-olds, respectively, required dental treatment and that no details of any bone grafting were available for 15 per cent of the 12-year-olds. This together with widespread evidence of poor record keeping and inadequate databases suggests that a reorganization of cleft care is now needed. There are few, if any, studies where an independent research team has been provided with access to patients and records to assess care and outcomes. Therefore, comparisons are difficult, but it says much for the professionalism of those involved in cleft care in the United Kingdom who enabled this study to take place. The Eurocleft Study (Shaw *et al.*, 1992) provides some data which reinforces the view that cleft care is in need of reorganization. The two U.K. Centres involved in the Eurocleft study had poor dental arch relations which, from the data pro-



vided here, is representative of the U.K. The current study examined patients born 10 years after those in the Euro-cleft Study and it seems that little has changed over this 10-year period. Data from Oslo on alveolar grafting, again from an historical and large dataset, showed that this technique has a failure rate of 4 per cent (Bergland *et al.*, 1986). The unacceptably high morbidity of this technique (42 per cent) in the U.K. suggests a serious reappraisal is needed. Interestingly, the failure rates for this technique were not significantly different between surgeons trained in the Plastic Surgery, and Oral and Maxillofacial Surgery specialities. The surgical, speech, and hearing data will be reported in appropriate and relevant Journals, but it is clear that all disciplines involved in cleft care need to reconsider their approaches in providing treatment. The use of neonatal appliances in nearly half the children of both age cohorts was surprising. A meta-analysis of the literature in this area is not possible because of the paucity of sound scientific studies, but the technique has never delivered any measurable or proven benefit, despite powerful proponents. The current study was not designed to measure their benefit since this requires prospective randomized clinical trials. There is at least one such study in progress and preliminary results indicate that the added cost and burden of neonatal appliances appears to confer so, benefit to speech, feeding, and maxillary arch dimensions (Kuijpers-Jagtman and Prahl-Andersen, 1997).

One major issue that has not been resolved by this study is the relationship between the volume of the unit and outcome. Elsewhere it has been suggested that a minimum caseload is required to maintain competence and proficiency, not only for surgery but also for orthodontics (Shaw *et al.*, 1996). There are, in addition, arguments for treatment of significant numbers in a unit since without this meaningful audit is impossible. On average, the majority of the 75 surgeons operated on less than one unilateral cleft lip and palate case per year. Since there were 105 orthodontists involved in cleft care it is not likely that the majority treat a significant volume of these patients. The lack of high volume operators and overall poor standard of results precluded a thorough analysis of the relation between outcome and volume in this study.

The widespread appreciation of care received by parents and children is similar to previous studies in the U.K. (Thomas *et al.*, 1997; Turner *et al.*, 1997). The area of most concern for parents and children was the appearance of the teeth. This is not totally unexpected since definitive orthodontics for a child with a cleft is usually completed after twelve years of age. The appearance of teeth and associated teasing causes the greatest distress of all facial features in non-cleft populations (Shaw *et al.*, 1980). It is hardly surprising then that teeth were an area of dissatisfaction. Satisfaction with care and outcomes is always difficult to measure objectively. There is also some evidence that the satisfaction with appearance in cleft subjects alters with age (Broder *et al.*, 1992; Thomas *et al.*, 1997). The present study represents only another cross-sectional view and, currently, true longitudinal data is lacking. Comparative data for parent satisfaction with cleft care treatment is available. Turner *et al.* (1997) in a nine-centre study in the U.K. found over 85 per cent of parents and patients were satisfied with their cleft care treatment, despite problems such as the missed diagnosis of cleft

palate. Reasons for this high satisfaction even with poor outcomes have been explored previously. These might include an indication of the trust of parents to cleft teams. Having entrusted their child's care, as well as investing emotional and physical effort during treatment, any complaint would be perceived as not justifying their own efforts. Finally, respect for professionals and possible intimidation within a clinical environment may lead to underestimation or under reporting of dissatisfaction.

The training of orthodontists involved in cleft care would benefit from a review. The areas of main concern in this study were the lack of involvement in cleft audit projects of recently appointed consultants. Without audit the incremental improvements in care and outcomes are not likely to occur. Both the newly-appointed and senior orthodontic consultants felt training would be beneficial in high volume centres, and this agrees with the final CSAG recommendations to the Minister of Health.

Clearly, the recommendations have implications for orthodontists and perhaps the main issue is that a centralized service would require involvement of fewer consultant orthodontists. Although a centralized service may be desirable to improve outcomes it does limit access for patients. Arrangements on a 'hub and spoke' service may overcome this problem (Shaw *et al.*, 1996). Such a system must envisage additional training for 'spoke' staff but guidance from, and regular visits to, the 'hub' specialists. This is workable within the current NHS system, but would need some reorganization. With 'hub and spoke' services the core team and specialist equipment are located at the 'hub', and this also becomes the co-ordinating centre for keeping records and implementation of audit. These arrangements also have implications for the training and continuing professional development of cleft clinicians who contribute to cleft care. There is a need to train future specialist clinicians only in sufficient numbers to match future posts at centres with a high case load and good clinical outcomes with commitment to audit. The current training of orthodontists was surprising in that some who had recently been appointed to cleft teams had no experience in basic procedures, such as the taking of neonatal impressions or the provision of definitive orthodontic treatment for patients with clefts. The CSAG Committee has suggested that future consultants involved in orthodontic care for children with cleft lip and palate should have a formalized training extending over at least 2 years. This training must include all aspects of orthodontic support for cleft palate treatment including supervised experience in orthodontics prior to alveolar bone grafting and orthognathic surgery. These consultants must also have sufficient experience to be involved in multi-centre studies on appointment, and this will only be achieved if audit and research experience are also acquired during training.

Orthodontists are often pivotal in record collection within a unit. Recommendations for minimum data sets were made by the CSAG Committee and it is essential that all clinicians involved in cleft care should make sure that the information is obtained for every child. It is also essential that this information is available so that comparisons of outcomes can be made both within the U.K. and with external centres.

One other implication of centralisation is that patients will have to travel greater distances for better care and that the additional financial burden on those families may need to be re-imbursed. The CSAG study did not carry out any objective assessment of the financial implications of centralising cleft care. Currently, this information is not available, but there may well be some economies of scale by sending all patients to a specialist centre. Such an approach avoids duplication of resources and, where costly equipment is involved, it is likely to be to its full capacity. This is also true for surgical and support staff where the numbers involved, including those in training, will be reduced. It must be emphasized that the recommendations made in the report were based not on the grounds of costs, but on the best interests of the patients. However the reduced need for re-operative and efficient orthodontic protocols represent considerable potential savings to the NHS

The next challenge is to respond to these recommendations and to meet the new training requirements, possibly through the development of newly accredited cleft training units. If the recommendations are not implemented it is doubtful if resources will be available to conduct a further survey in 10 years time. Furthermore, if there is no change it is unlikely that outcomes would be significantly different to those in this report. It appears inevitable that children born with a cleft lip and palate can only expect poor outcomes if the current organization of care continues.

### Acknowledgements

This study was only possible because the enormous additional burden of the project placed on all participating cleft teams was met in full. The authors acknowledge all their commitment to these children with thanks and admiration. We are grateful for the guidance and useful discussions with all of the members of the CSAG Cleft Lip and Palate Committee. Computer support was cheerfully provided by Phil Eden, Tremaine Cornish and Arvin Prasher; Andy Leach and Bob Cappelli produced hundreds of models, Mike Mars, Nikki Atack, and Gunvor Semb generously gave their time to analyse models and X-rays. Helpful advice was given by the CSAG Support Team in Oxford, particularly Dr John Newton and Dr Elizabeth West. Jane Western, Janice Richardson and Roger Motton were responsible for photocopying and typing thousands of pages of datasheets and reports. We are grateful to the Department of Health for funding this project, and finally, we thank all those children and parents who willingly participated in this study.

### References

- Atack, N., Hathorn, I., Mars, M. and Sandy J. (1997)**  
Study models of 5 year old children as predictors of surgical outcome in unilateral cleft lip and palate,  
*European Journal of Orthodontics*, **19**, 165–170.
- Bergland, O., Semb, G. and Abyholm, F. E. (1986)**  
Elimination of the residual alveolar cleft by secondary bone grafting and subsequent orthodontic treatment,  
*Cleft Palate Journal*, **23**, 175–205.
- Broder, H. L., Smith, F. B. and Strauss, R. P. (1992)**  
Habilitation of patients with clefts: parent and child ratings of satisfaction with appearance and speech,  
*Cleft Palate and Craniofacial Journal*, **29**, 262–267.
- Jones, M. L. (1996)**  
CLP, CSAG and all that!—the testing of a hypothesis,  
*British Journal of Orthodontics*, **23**, 163–164.
- Kuijpers-Jagtman, A. M. and Prah-Andersen, B. (1997)**  
Value of presurgical infant orthopaedics: an intercentre randomized clinical trial,  
In: *Transactions of the Eighth International Congress on Cleft Palate and Related Craniofacial Anomalies*,  
Stamford Press, Singapore, p.1002.
- Mars, M., Plint, D.A., Houston, W. J. B., Bergland, O. and Semb, G. (1987)**  
The Goslon Yardstick: a new system of assessing dental arch relationships in children with unilateral clefts of the lip and palate,  
*Cleft Palate Journal*, **24**, 314–322.
- Mølsted, K., Asher-McDade, C., Brattström, V., Dahl, E., Mars, M., McWilliam, J. Plint, D. A., Prah-Andersen, B., Semb, G., Shaw, W. C. and The, R. P. S. (1992)**  
A six-center international study of treatment outcome in patients with clefts of the lip and palate: Part 2. Craniofacial form and soft tissue profile,  
*Cleft Palate Craniofacial Journal*, **29**, 398–404.
- Shaw, W. C., *et al.* (1992)**  
A six-center international study of treatment outcome in patients with clefts of the lip and palate,  
*Cleft Palate Craniofacial Journal*, parts 1–5, **29**, 393–418.
- Shaw, W. C., Meek, S. C. and Jones, D. C. (1980)**  
Nicknames, teasing, harassment and the salience of dental features among school children,  
*British Journal of Orthodontics*, **7**, 75–80.
- Shaw, W. C., Williams, A. C., Sandy, J. R. and Devlin, H. B. (1996)**  
Minimum standards for the management of cleft lip and palate: efforts to close the audit loop,  
*Annals of the Royal College of Surgeons*, **78**, 110–114.
- Thomas, P., Turner, S. R., Rumsey, N., Dowell, T. and Sandy J. R. (1997)**  
Satisfaction with facial appearance among cleft affected subjects,  
*Cleft Palate-Craniofacial Journal*, **34**, 226–231.
- Turner, S. R., Thomas, P. W. N., Dowell, T., Rumsey, N. and Sandy, J. R. (1997)**  
Psychological outcomes amongst cleft patients and their families,  
*British Journal of Plastic Surgery*, **50**, 1–9.
- Williams A. C., Shaw W. C., Sandy J. R. and Devlin H. B. (1996)**  
Consultant Orthodontic Services for Cleft Patients in England and Wales,  
*British Journal Orthodontics*, **23**, 165–169.