Do You Care? A National Register for Cleft Lip and Palate Patients

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Abstract: *The Cleft Palate Index and, more recently, the Craniofacial Anomalies Register—CARE—have been in operation since 1982. This paper summarizes its development and plans for the future. CARE is a multidisciplinary committee involving all specialities involved in the treatment of this group of patients therefore it should and can be well placed to co-ordinate the cleft data arising from these patients.*

Development

CARE was instigated as a data registry for all specialities involved in cleft care. The founding members of the Cleft Palate Index were elected at a Craniofacial Society meeting in 1980. Representatives were elected from the four main Specialities involved in Cleft care—Orthodontics, Plastic Surgery, Maxillofacial Surgery and Speech and Language Therapy. Input forms were subsequently designed and the Central Cleft Palate Index was established in 1982 with 30 participating units. Data was collected under the auspices of the Craniofacial Society. CARE is a Standing committee of the Craniofacial Society. Improvements to the methods of data collection were made and the group was renamed the Craniofacial Anomalies Register (CARE). Revised input forms were issued called 'CP1reg' to record birth details and 'CP1surg' to record details of operative procedures. Another arm of CARE has been concerned with the development of standards. CARE has been funded by contributions from the Craniofacial Society, British Orthodontic Society, British Association of Oral & Maxillofacial Surgery and the British Association of Plastic Surgeons.

Why Collect Data

There are several important reasons why we should collect data.

1. *Research and audit*—There are a small number of cleft patients born in each Health District, so pooling of data will in the future produce a larger resource for research, often making statistical significance greater. The emerging possibilities offered by clinical and molecular genetics require a clearly defined family genetic pedigree, to assess effectiveness of any new treatments.
2. *Audit*—The recording of consecutive cases of a condition helps in preparing a series of patients with similar cleft deformities and allows comparison for audit purposes, in order to assess the effectiveness of treatment and set standards for the future.
3. *Purchasing and planning cleft services*—Recording the incidence of birth deformities allows for the planning and provision of services to cater for these children. It has generally been regarded that the incidence of cleft subtypes in the United Kingdom is about 1:700 [Clinical Standards Advisory Group (CSAG), 1998]. It is important to have accurate figures for the number of children requiring treatment to perform a population-based needs assessment.
4. CARE records cleft registrations against the hospitals and cleft teams that treat these cases. It therefore has information about cleft teams and workload that is useful in planning training centres for future cleft team members.

How CARE Works

The 'Caretaker' registers the patient on the 'registration form' with the CARE database. Caretakers are either a member of a Cleft team at the 'hub' or may be at a 'spoke' to the Cleft team. This wide coverage helps to ensure that as many cleft babies are registered as possible (see Figure 1). Children with clefts are usually registered in the first year of life. The registration form is designed to collect those details usually obtained at an early consultation with the parents. However, the peak registrations of cleft patients is not for 3–4 years, as some of the less obvious cleft types are at first not noticed or diagnosed, and may only become apparent when speech problems develop. The registration form is a 4-page 'no carbon required' form. One copy is sent to the CARE co-ordinator to place the patient on the CARE database; another goes to the Regional co-ordinator, and one copy is retained in the patient's hospital notes or retained by the Caretaker for their own records. The last sheet is given to the parents so that they can understand and know what information is held on the database. It is also hoped that the parents will contact CARE with any amendments if the information changes.

A network of Regional co-ordinators has been established by Iain Hathorn. The Regional co-ordinator's role is two-fold. First, they have local knowledge of the cleft teams operating in their Region, and act as a link between the

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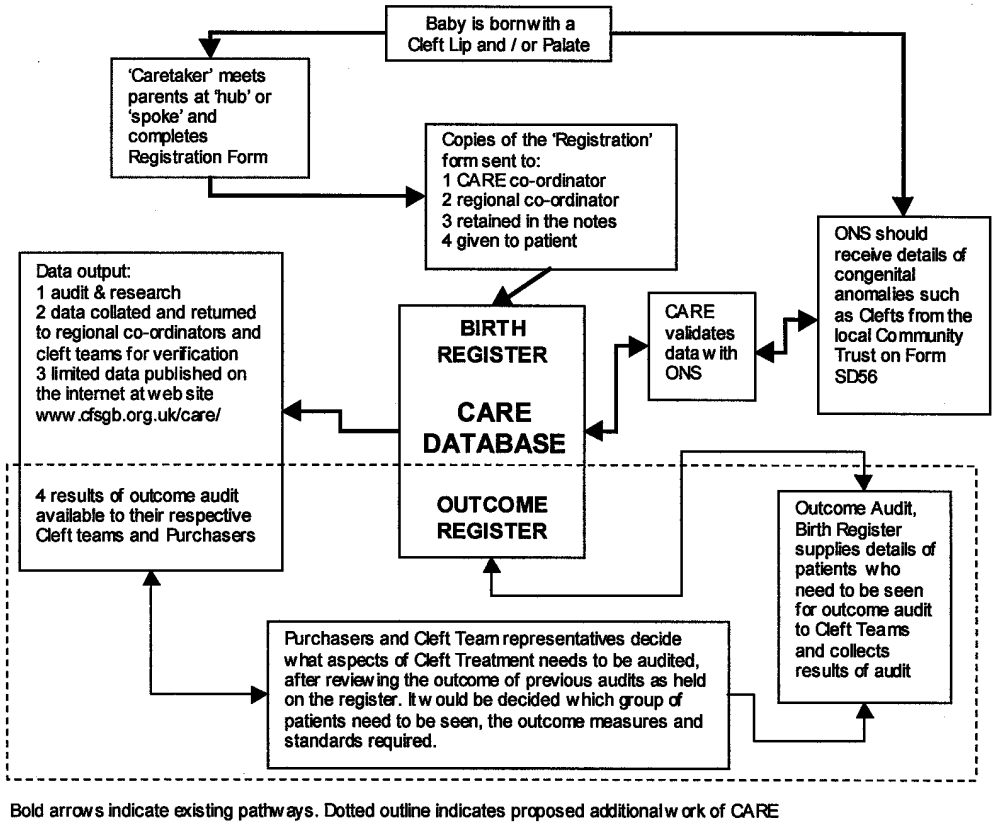

FIG. 1 Organisation of CARE.

Coordinator	Mark Hammond	Orthodontics
Deputy Coordinator	Leo Stassen	Maxillo-Facial Surgery
	Tim Goodacre	Plastic Surgery
	Roz Razzell	Speech & Language Therapy
	Peter Gornal	'Other Specialities' (Paediatric Surgery)
	Iain Hathorn	Orthodontics
	Mike Dixon	Genetics

FIG. 2 CARE Committee.

CARE committee and the Caretaker's and cleft teams. Secondly, they also act as 'motivators' to maintain a flow of registrations from the Cleft teams in their Region. Estimates of the number of potential cleft subtypes, in each Region, are calculated from the number of births in each region (data supplied by ONS). Caretakers and regional co-ordinators have access to web pages on the internet. These pages show the estimated number of cleft sub-types and the number of patients actually registered with CARE both by Region. The Regional co-ordinator can then assess how completely the data is being collected in their Region. The information provided on the Internet is anonymised and simply quantitative.

The CARE committee is composed of one member from each of the main Specialities involved in the management of these patients, (Figure 2). Another member is elected from the 'Other Specialities' section of the Craniofacial Society. A further member is elected to give advice in the field of clinical genetics. Election of the co-ordinator revolves between the main Specialities, over an agreed term of office. The Deputy Coordinator becomes the Coordinator in due course.

The CARE co-ordinator oversees the operation of the regional co-ordinators and maintains the CARE database with the help of an elected committee. The Co-ordinator liaises with ONS through their BINOCAR initiative (British Isles Network of Congenital Anomaly Registers) to validate the data and produce more accurate data for ONS to disseminate. The CARE co-ordinator receives requests for data. At the inception of CARE, a confidentiality barrier was used centrally so that the information released would only be with the approval of the local Caretaker and team. A request for data would be forwarded to the units where patient records are held to obtain their approval before data is released. This method of data output was thought to be most appropriate to protect

CARE - Craniofacial Anomalies REgister - Registration Form

1 - CARETAKER

Please PRINT and use a sticker from the Hospital notes where possible. Read the form from right to left

Caretaker's name and designation			
Patient's home NHS Region		Regional Co-ordinator	
Cleft Centre		Cleft Centre No.	
Hospital - if not Cleft Centre		Hospital No.	

2 - PATIENT

Surname & Forename(s)			
Previous name (if any)			
Street/District			
Town/City		County	
Postcode		Date of Birth	
Gender	M F	NHS No.	

Sections numbered 1-3 provide us with the essential information required; and it helps CARE if you complete the form as fully as possible.

3 - CLEFT DETAILS

	Right						Left					
	patient's side											
Simonart's Bands	Y											Y
Lip		I	C						I	C		
Alveolus			I	C				I	C			
Hard Palate					I	C						
Soft Palate					I	C						
Cleft Summary												

I=Incomplete C=Complete

Vomer attached to Hard Palate	Y	N
Submucous Cleft	Y	N
No Cleft, but seen for VPI	Y	N

If cleft is part of a syndrome, give its name | Pierre Robin | Y | N

4 - PREGNANCY

Birth at Home	Y	N	Or name of Hospital and town/city							
Mothers Hospital No.			Birth Weight		Gestation (Full Term=40)					
Pre-natal diagnosis?	Y	N	Scanned at week		History of miscarriages	Y	N			
Antenatal History of	Smoking	Y	N	Alcohol	Y	N	Drug Abuse	Y	N	
Details or other findings	Medication	Y	N	Vitamins	Y	N	Operations	Y	N	
	Folic Acid	Y	N	Illness	Y	N	Trauma	Y	N	
				CLAPA / support group contacted				Y	N	

5 - MED HIST

Medical History- any abnormalities of			GMP			GDP		
Cardiovascular system	Y	N	Endocrine system	Y	N	Bleeding diathesis	Y	N
Renal system	Y	N	Skeletal system	Y	N	Craniofacial area	Y	N
Respiratory system	Y	N	Eye abnormalities	Y	N	Regular Medication	Y	N
Details or other findings								

6 - MUM

Date of Birth		Occupation					
Ethnicity	White	Black Caribbean	Black African	Indian	Pakistani	Chinese	Specify
				Bangladeshi			
First Language		English	Y	N	Other		

7 - DAD

Date of Birth		Occupation					
Ethnicity	White	Black Caribbean	Black African	Indian	Pakistani	Chinese	Specify
				Bangladeshi			
First Language		English	Y	N	Other		

8 - GENETIC

Does Mum have a cleft ?	Y	N	Maternal family history of clefts			Y	N	
Does Dad have a cleft ?	Y	N	Paternal family history of clefts			Y	N	
Number of **other** children **with** clefts	M		F					
Number of **other** children **without** clefts	M		F					
Twins Y N	Consanguinity	Y	N	Agree to blood sampling			Y	N
Referred to geneticist?	Y	N	Name of Geneticist					

Sep. - 1998

Return Registration forms regularly, rather than returning a large batch periodically. White copy to Dr. M Hammond, Consultant Orthodontist, Dept. of Orthodontics, Carbett Hospital, Stourbridge, West Midlands, DY8 4JB. Pink copy to the regional co-ordinator. Yellow copy place in the patient's notes or keep yourself. Please hand the blue copy to the parents. If the parents later find that the information changes please ask them to send details to Dr. M Hammond at the above address.

FIG. 3.

locally sensitive information and obtain the co-operation of cleft teams. Nearly all units now find this unnecessary and have withdrawn the need for a confidentiality barrier. However, some University departments who have been recording information about patients still like to protect this data and have expressed some reservations about the availability of their data.

The Data Collected So Far

CARE at present collects data for England and Wales. CARE has been largely concerned with descriptive data. This was seen as a useful first step in the epidemiology of this condition to provide an overview of clefts. The original intention in collecting data was that it would act as a signpost to locate further patient details, rather than be an exhaustive data set. Consequently, the early datasets were minimal. They collected data on cleft type, Hospital number and the presence or absence of 'other abnormalities'. In an increasingly busy working life it has become important to balance the needs for a useful dataset against the time involved in completing the registration form and entering the data on the database. The form now in use is shown in Figure 3. Looking to the future multicentre audit may well involve European centres more frequently. We will liaise with European Biomed and ensure our datasets are compatible with theirs.

The value of a national database is to provide descriptive data for cleft teams, to show changes in the incidence of the condition and to maintain surveillance for increases in the incidence of clefting. To help ensure the accuracy of this data, cases registered since 1995 are being validated against data held by the Office for National Statistics (ONS).

CARE has been more successful in recording cleft cases then ONS (Williamson personal communication) Figure 4. The true number of cases being seen is important to record as the planning and provision of services by Public Health departments is usually based around ONS data. Recording data of this type is beset with difficulties. Inaccuracies may arise from simply being unable to read someone's handwriting, inaccurate data input and, in many cases, the Caretaker thinking they have registered a patient when they have not. The only way to improve the accuracy of the data is to regularly send lists of the patients registered on the database together with their details, so that the local team can verify the accuracy of the data and correct any mistakes.

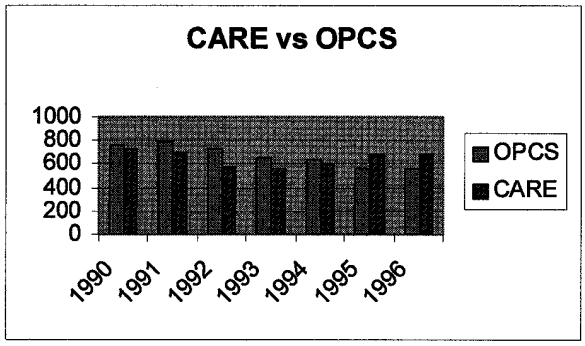

FIG. 4.

Form CP1surg tried to capture information about operative procedures and dates of operations. However, the infinite number of variations on the basic operative procedures make it difficult to compare one centre's operative procedure with another. In future, it is hoped to record information that will be more useful for audit. Therefore, we will still record dates of the primary lip and palate operations, and some secondary procedures, but will not record details of operative procedures. This should allow us to produce lists of consecutive operations for retrospective audit purposes. It is increasingly important to record outcome audit data in addition to registration data to provide useful information for Cleft teams. This is likely to be a main theme for CARE in future years.

Requests for data from CARE are encouraged. Data can be despatched quickly if only an anonymised response is required. If data are likely to identify patients or a particular Cleft team, then the request is either discussed by the CARE committee or is forwarded to the centres involved to ask their permission to release the data.

How Complete is the Data?

The committee at CARE appreciates that the database is not complete, because not all patients are registered. Voluntary reporting of clefts is not ideal and until reporting becomes mandatory the database is unlikely to hold all the patients. Therefore, other ways of obtaining patient details are being sought to remedy this. Details of these patients can then be transferred to the CARE database. Two ways used are:

1. *Other databases*: ascertainment of congenital anomalies arose from the Thalidomide disaster of the 1960s. However, today only in Denmark is the reporting of facial clefts compulsory, where, the incidence is reported to be 1:529 live born infants in 1981 (Jensen *et al.* 1988). The Office for National Statistics (formed on April 1st 1996 by the merger of the Central Statistical Office, and the Office for Population Censuses and Surveys), is a government agency for compiling, analysing, and disseminating many of the United Kingdom's economic, social, and demographic statistics. It collects congenital anomaly statistics for England and Wales on the National Congenital Anomalies System register. ONS receives data about congenital malformations by completion of form SD56 locally at the place of birth. Form SD56 is completed mainly by Physicians and Midwives, with data supplemented by reports from Neonatal and Special Care Baby Units. Community Trusts forward this information to ONS on behalf of the District Health Authorities. Reporting anomalies through this system is again voluntary. Data is stored using the International Statistical Classification of Diseases and Related health Problems, 1992 (ICD 10). Congenital Anomaly Statistics notifications are published every two years by ONS. The most recent report covers the years 1995 and 1996. ONS has formed the BINOCAR initiative (British Isles Network of Congenital Anomaly Registers) to pool data from registers such as CARE that exist throughout the Country.

   The Office for National Statistics contributes information to the International Clearing House for Birth

Defects Monitoring Systems. This monitoring programme, based in Rome, was started in 1964 and was instigated as a surveillance system to record congenital anomalies. Significant changes in incidence of a congenital anomaly are reported to the local District Health Authority. Significant changes do not only occur because a rise in birth incidence but also through better diagnosis and reporting as may occur when a new Midwife or Physician starts to work in an area. It is therefore important that ONS collaborates with others to ensure the data is accurate. ONS also contributes data to the European Registration of Congenital Anomalies Register (EUROCAT).

In the United Kingdom reporting is on a voluntary basis so it is important for CARE to liaise with ONS to improve the accuracy of the data. The data collected by ONS is not sufficiently detailed for prospective clinical audit and research so CARE data and ONS data should be used to complement each other.

2. The second method is to obtain data held by the Regional outposts of the NHS relating to operation codes for primary lip and palate surgery. Outposts hold data on hospital number, date of birth, district code, and type of operation. The data relating to operation codes for primary lip and palate is sourced. Details of patients who appear to have had primary repairs are noted and the local team is contacted for further details so they can be entered on the database. However, not all Regional outposts are able to supply this data.

Complete registration of all cleft patients is unlikely to occur unless:

1. Reporting is made compulsory through legislation.
2. Data clerks are available to help collect data.
3. Efficient forms are used that are not too elaborate and can be completed accurately without ambiguity. Some centres have already combined their basic stationery, for example discharge letters, with the details required by CARE.
4. Cleft teams are confident that the information will not be used to undermine their work. This requires strict protocols for data input and output.
5. Some units will require help in setting up systems to capture the data.

A National Register for Outcome Data

It is hoped that CARE, following wide discussion, can record outcome audit data and put in place a mechanism for external audit. Consequently, CARE will become a national register of both cleft patients and their treatment outcomes. It is important to know what outcomes measures are being achieved to set appropriate standards and aid protocol setting for the future. CARE will review with Cleft teams the outcome data and help set up regular reviews of this data to improve treatment in all centres. These standards should be tested by regular externally assessed audit (Williams & Markus, 1998). CARE will discuss with cleft teams and purchasers what are the appropriate outcome measures to record.

National registers already exist for many clinical conditions, for example, the United Kingdom Children's Cancer Study Group (UKCCSG), which provides support to programmes of research. The UKCCSG is fed data through a network of 22 regional assistants. In some clinical conditions the outcome is clear-cut, for instance survival times following diagnosis. Registers can record theses survival times and then put in place reviews of services not meeting the currently acceptable standards. Clearly, in the treatment of cleft patients the outcomes are more complex and discussions need to be held to determine the outcome criteria.

Recent reforms in the National Health Service following the consultative document 'The new NHS—Commissioning Specialised Services' and the White Paper 'The New NHS' clearly point to new commissioning mechanisms for specialist services, such as Cleft Lip & Palate, through the Regional Specialised Commissioning Groups (RSCG). The RCSG will set targets for cleft teams in the future. To set these targets they will need comparative data. CARE should play a vital role in collecting and disseminating this data because of its multidisciplinary nature. Comparative information and target setting (contracting) will be in the public domain as it is with other services. For example, data comparing survival times following cancer diagnosis and treatment are already collected, and are being used to compare and improve teams. Comparative data and 'league tables' already exist in Cleft services, for instance, those in Eurocleft and the CSAG reports. These may be anonymized, but they do show the range of results that are being achieved. Purchasers will be looking to improve the work of all cleft teams and especially the less good, to deliver quality treatment. The RSCG will require information on audit and cleft teams to help in planning services.

Another arm of the recent reforms is performance monitoring and this is to be introduced into clinical areas through the evidence-based National Service Frameworks, inspired by the Calman-Hine Cancer Report 1995. The consultation document outlines standards of service and availability of services. Information will be collected and disseminated about individual performances. Medical directors will be charged with ensuring that the trusts data meets nationally agreed national standards. The National Institute for Clinical Excellence (NICE) will be set up to provide information about both clinical and cost effectiveness. It will produce and disseminate clinical guidelines with linked information on clinical audit and will bring together work currently undertaken by different organisations. CARE is well placed to present information and offer advice to these new agencies.

The Future

Following the publication of the CSAG report and discussions with cleft teams CARE wishes to pursue the following.

1. The strength of CARE is in its network of participants that cover the majority of England and Wales. It is hoped to encourage those centres not presently contributing to CARE to enter CARE data in future.
2. The Craniofacial Society contributes to the funding of CARE but has no role in its management. A report is sent to the funding organizations annually. If data are

to be collected more accurately than at present this will involve greater resources. We will lobby the government through groups such as the CSAG Implementation Group to fund CARE's activities through contracts with the RSCG. It may then be appropriate to consider developing CARE as a separate identity, independent of the Craniofacial Society, with a new constitution.

3. It is hoped that CARE will act as an umbrella organisation for the collection of cleft lip and palate data for the United Kingdom, making this information available to all concerned with the care of these patients. We are collaborating with Welsh, Northern Ireland and Scottish (SCALP) databases to ensure the datasets are comparable and that data is exchangeable.
4. Until reporting of clefts becomes compulsory, CARE will work with the Office for National Statistics to produce as accurate incidence rates of cleft types as possible. We will lobby the government to make recording of congenital anomalies compulsory.
5. Output from any database is essential, otherwise the data are redundant. CARE will encourage greater use of the data. A World Wide Web site has been established to publicise the work of CARE and publish a limited subset of data for Cleft teams information. This can be viewed at 'www.cfsgb.org.uk/care/'. It is hoped to publish more data in the future with the approval of the contributing centres and when the data is acceptably accurate.
6. CARE will prompt for the collection of clinical records, so that regular externally assessed audit can be accomplished more readily. How external audit is to be organized is subject to discussion. However, if the necessary records are available it should make the work easier. We will use the recommendations of the Royal College 1995 as a basis for record keeping. Form CP1surg will be redrafted to record basic treatment details and as a prompt for record collection.
7. Perhaps the greatest challenge for the future is recording outcome data in a meaningful and accurate way. The establishment of a National Register of outcome data, we hope, will be a priority for cleft teams. This has to be done in a non threatening way to ensure compliance. It is hoped that the Implementation Group will see this as a priority and recommend funding a multidisciplinary group such as CARE through contracting to carry this out.

The CARE database is not a clinical management tool for individual units. Software of this nature already exists. CARE does not have the resources to develop and field test such a program. Each centre will have a different wish for properties in such a software program which makes the development of such a tool expensive. Obviously, the core datasets in such a program should be compatible with CARE to avoid double entry of data and CARE is happy to work with software programmers to achieve this.

The committee looks forward to working with cleft teams to improve data collection in the future. Data collection is not easy! The committee appreciates everyone's time and patience to collect the data and ensure it is accurate. Please return forms to us steadily throughout the year. On our part we will send you information that is on the database regularly.

If you are interested in joining CARE please contact Mark Hammond

References

Clinical Standards Advisory Group (1998)
Cleft Lip and/or Palate
London, The Stationery Office.

HMSO (1995)
A Policy Framework for Commissioning Cancer Services. Report of the Chief Medical Officers of England and Wales,
London, The Stationery Office.

HMSO (1998)
Congenital Anomaly Statistics Notifications Series MB3 No. 11 (1998). Office for National Statistics.
London, The Stationery Office.

Jensen, B. L., Kreiborg, S., Dahl, E. and Fogh-Anderson, P. (1988)
Cleft lip palate in Denmark, 1976–1981. Epidemiology, variability, and early somatic development.
Cleft Palate Journal, **25**, 258–269.

Royal College of Surgeons of England Surgical Audit Unit Cleft Lip and Palate Steering Group (1995)
Minimum Standards for the Care of Children with Clefts of the Lip and/or Palate.

Williams, J. J. and Markus, A. F. (1998)
Cleft Care: life after CSAG, Editorial,
British Journal of Oral and Maxillofacial Surgery, **36,** 81–83.