

Editorial**Improving services for cleft lip and palate. A work in progress**

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Services and treatment protocols for the management of children with cleft lip and palate can be remarkably different within and between countries. In Europe, there is wide variation from poorly coordinated disparate services, as in Portugal or Greece, to highly centralised specialised teams as in Scandinavia. Clinical uncertainty is evident from a striking diversity of practices. In one survey, 201 European teams carrying out primary surgical repair for unilateral clefts, employed 194 different protocols, reflecting the paucity of published randomised trials in the field (Shaw *et al*, 2001).

A series of WHO consensus conferences between 2000 and 2005 confirmed that service organisation and treatment uncertainty are widespread international problems, while lack of resources puts the most basic surgical treatment beyond the reach of thousands of children in the developing world (WHO, 2002).

Regional Perspectives

Representatives of several countries and regions provided the following international overview at a consensus conference in 2005:

In sub-Saharan Africa clinical resources for CFA are scarce as a consequence of prevailing economic problems and the greater challenge of communicable diseases, particularly AIDS. For example, in Namibia despite a high reported incidence of clefts, there are no cleft surgeons. As the wealthiest sub-Saharan country, South Africa has around 12 centres that undertake cleft surgery but these tend to work independently without common quality-improvement protocols and there has been little formal study of clefting in the African population. There are a number of centres in the cities of Northern Africa but, as elsewhere in Africa, a survey has yet to be undertaken to identify potential sites with capability for collaborative research.

In Australia and New Zealand there are well-developed services in many cities, though in some instances, the case-load is quite low, limiting the potential for collaborative research and arguably, high volume expertise. However, the establishment of the Australian and New Zealand Craniofacial Association makes coordination possible and one centre has a programme of support and development for Indonesian and Malaysian cleft centres.

In China there is a high level of unmet need for individuals with clefts. Treatment is not free and follow-up is difficult. Speech therapists are especially scarce.

There is, however, a network of several large surgical centres that could form a potential research partnership.

European clinical services were surveyed around a decade ago (Shaw *et al*, 2001), since which there has been little change with the exception the UK (Bearn *et al*, 2001). In the main, Europe's problems arise from fragmentation of care over numerous small centres. Scandinavia, especially Denmark, Norway, and Finland is the exception, with a small number of long-established, well resourced, national centres of excellence.

The Indian subcontinent is yet to be surveyed regarding cleft services and research capability. However, an overview of India was presented and may be reasonably representative of adjoining countries. There are high levels of unmet need and access is complicated as the majority of the population live in rural communities. There are several hundred surgeons trained in cleft surgery and several large university hospitals but, as yet, no quality-improvement protocols are in place. The subcontinent undoubtedly has numerous potential partners for clinical trials though resourcing follow-up studies will be a challenge.

No survey has been undertaken on clinical services and research capability across Latin America and the Caribbean. Mexico was represented at the WHO conference and has at least one large centre in Mexico City that has successfully completed clinical trials, and is recognized as a centre of excellence in the region. Brazil was also represented by a large centre of excellence at Bauru. Elsewhere in Latin America there is likely to be a high level of unmet need.

In South-East Asia, Singapore has well developed services and there is a large centre of excellence in Taipei. In Indonesia there are high levels of unmet need but around six cleft teams are established and would be potential sites for research collaboration. Already both Indonesia and Malaysia are engaged in epidemiological, nutritional and genetic research with agencies in Australia, Europe, Singapore and elsewhere. Like Europe, Japan suffers fragmentation of services in small centres, however, the Japanese Cleft Palate Association has begun discussions on intercentre studies and clinical trials. In Korea, several high-volume centres are potential sites for collaborative research and the Korean Cleft Palate Association has begun discussion on intercentre studies. Thailand has also been developing services and has an increasing capability for research.

In the Middle East, a high level of unmet need has been reported with few established cleft centres.

North America also suffers from a fragmentation of cleft and craniofacial services, and representatives from the USA reported little possibility of obtaining sufficient subjects for clinical trials because of the decentralized nature of services. The emergence of health management organizations over recent decades was seen as a particular force for the demise of established cleft teams. None the less, the American Cleft Palate-Craniofacial Association continues to promote team care and has published several sets of guidelines.

The absence of local services in the poorest countries is partly met by an increasing number of non-governmental organisations such as Operation Smile, Smile Train, and many others. These voluntary groups provide aid in a variety of ways, from short-term visits of foreign operating teams to the provision of financial incentives designed to encourage local hospitals to undertake more cleft surgery. As yet there is little formal international governance of these activities and the quality of NGO contributions ranges from transient and sometimes incompetent intervention to substantial lasting investment in local infrastructure and training. Ironically, the USA and Europe have also been exporting complex, expensive, and burdensome protocols to developing countries. These include varieties of presurgical orthopaedic treatments that have yet to be critically evaluated and are of dubious worth.

Mixed Success In Improving Services

The Eurocleft cohort studies, started in the mid-1980s, were blinded intercentre comparisons of surgical outcome in consecutive matched cases from different institutions. Early findings indicated that outcomes could be substantially different between centres, simple protocols could match or surpass the results of complex high-burden protocols, and successful care was associated with centralised teams whose main work focussed on cleft treatment (Semb *et al*, 2005). One particular finding was the extremely poor standard of care in two UK centres, later confirmed as a systemic national problem. Government intervention subsequently centralised UK services on dedicated regional teams (Bearn *et al*, 2001).

Though a European network, developed with subsequent EU support produced consensual guidelines on team care in Europe, restructuring has been nowhere as radical as in the UK. Interestingly, the obstacles for those seeking change elsewhere in Europe were mainly non-financial. Instead the difficulties reported were:

- Personal egotism of individual practitioners unwilling to discontinue the practice of treating a few children each year;
- Competition between specialties for pre-eminence in the field, e.g. plastic-vs-maxillofacial-vs-paediatric-vs-ENT surgery;
- Local pride, with every hospital, town or region desiring its own small team or wishing to have local service;
- The desire of teaching hospitals to cover a spectrum of clinical practice for junior doctors,
- Lack of responsiveness in the health authorities at local and national level.

Indeed it is probable that the same obstacles in the UK would have impeded change had there not been government intervention.

However, many centres in Europe have now begun to participate in blinded intercentre comparisons, and this may lead to wider progress over time. Furthermore, successful intercentre collaborations are becoming established elsewhere, such as “Americleft”, “Japanleft”, and “Indiacleft”.

Mixed Success In Improving The Evidence Base

One final consequence of the original Eurocleft cohort studies was to promote among participants, the transition from retrospective cohort study to randomized trials. One Dutch trial of presurgical orthopaedics has now been completed, and demonstrated a lack of worth for this ancillary procedure. The first three major international trials of primary surgery, the “Scandcleft” Trials, are in follow-up, and the fourth, a multicentre trial of the timing of palate surgery is underway.

Presently, however, such activities are confined to Scandinavia, the UK, and the Netherlands. Regrettably, some of Europe’s largest countries, Russia, Germany, France and Italy have yet to establish the networks that would permit a critical appraisal of services, and sustain collaborative cohort studies and trials.

Summary

Cleft lip and palate remains an orphan condition, falling between a variety of clinical disciplines, and often forgotten by public health consultants and health commissioners. Much remains to be done, and in the absence of leadership from clinicians or commissioners, progress may have to await the emergence of a stronger voice from organizations representing affected families.

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