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SYSTEMS CHANGE

Key challenges for hub and spoke models of care – A report from the 1st workshop of the EHC Think Tank on Hub and Spoke Treatment Models

Amanda Bok, Declan Noone, Naja Skouw-Rasmussen, on behalf of the EHC Think Tank

Introduction: The hub and spoke model can deliver high quality care to a scattered population through centres of expertise supported by a network of several smaller geographically dispersed centres. This approach is now being proposed to provide care for people with rare diseases, and in particular for rare bleeding disorders. To ensure that specialised treatments such as gene therapy can be delivered effectively using the hub and spoke model of care, it is important to understand the challenges that the model presents for all stakeholders. **Identifying key challenges:** As part of the EHC Think Tank Workstream on Hub and Spoke Treatment Models, 14 stakeholders representing health care providers, patient groups, research and industry met in November 2021 to identify challenges in the design, implementation and sustainable operation of hub and spoke models, and to propose ways in which resources could be allocated and collaboration fostered, from each of their stakeholder perspectives. Five key challenges were identified: 1. How future care might be re-envisioned; 2. Which agencies and stakeholders should determine which centres become hubs or spokes, and how this process might be carried out; 3. Identifying the criteria that will define a hub and spoke, and the roles of various stakeholders in that process; 4. How resources might be allocated; 5. How hubs and spokes

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It is important that the hub and spoke model continues to be effective in delivering high quality care for people with rare diseases. As gene therapy and other new specialised treatments become available, understanding the potential challenges associated with how the model will work is key.

AMANDA BOK
European Haemophilia Consortium, Brussels, Belgium

DECLAN NOONE
European Haemophilia Consortium, Brussels, Belgium

NAJA SKOUW-RASMUSSEN
European Haemophilia Consortium, Brussels, Belgium

EHC THINK TANK
European Haemophilia Consortium, Brussels, Belgium.
Email: office@ehc.eu

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will collaborate to ensure that patients' needs are prioritised. This model may also be recommended for treatment with gene therapy in certain rare diseases. Summary: Hub and spoke models should be implemented by establishing criteria for hub and spoke status, prioritising patients in service reorganisation and in the care pathway, and considering the impact of new service models on current arrangements. The next step is to vet the challenges identified by this workstream with a broader group of external stakeholders and bring their perspectives back for consideration.

Keywords: *Bleeding disorders, Rare diseases, Gene therapy, Health care facilities, manpower and services, Tertiary healthcare, Hub and spoke, Organisation of care, Long-term follow-up, Comprehensive care, Multidisciplinary team, Treatment centre networks, Data sharing, Patient pathways, Patient management*

A challenge common to the management of all rare diseases is how best to provide high quality care to a small number of patients who are geographically widely dispersed. This question has acquired new urgency as governments consider how to provide highly specialised expertise to deliver new high-cost technologies such as gene therapy for bleeding disorders and other rare diseases.

Clinical expertise is best supported by concentrating specialists in a few large centres that attract sufficient funding to provide high-level care [1-3]. However, the benefit for patients must be balanced by the barriers they face when accessing such a centre, which include geographical remoteness and the personal costs associated with attendance [4,5]. This balance is strongly favourable for geographically small countries or urban centres that have a large population and sufficient wealth to fund the service. It is less favourable where patients must travel long distances, in countries with a small population (and therefore few people with rare diseases), or where resources cannot sustain several large centres.

The hub and spoke model of care, in which a large centre provides specialist expertise to support many smaller geographically dispersed centres, is one way in which these conflicting needs can be addressed. The European Association for Haemophilia and Allied Disorders (EAHAD) and the European Haemophilia Consortium (EHC) have called for first-generation gene therapies to be introduced by means of a hub and spoke model whereby treatment is prescribed and managed exclusively by expert haemophilia centres (as the national hubs), and monitored by treatment centres in close communication with the primary expert hub (as spokes linking into that hub) [6]. To ensure that specialised treatments such as gene therapy can be delivered effectively using the hub and spoke model of care, it is important to understand the potential challenges associated with how the model will work.

IDENTIFYING KEY CHALLENGES

In the first session of the EHC Think Tank Workstream on Hub and Spoke Treatment Models in November 2021, 14 stakeholders representing healthcare providers, patient groups, research and the pharmaceutical industry participated in a virtual meeting to identify challenges in the design and implementation of hub and spoke models, and to

Figure 1. Template for identifying challenges for hub and spoke treatment models

Challenge:

For whom is this a challenge?

Why is it a challenge for them?

Who might hold a key to a solution?

Why?

propose ways in which resources could be allocated and collaboration fostered. Participants were split into four breakout groups to consider the key challenges in depth. Each group was tasked with developing a shortlist of three to four challenges based on a template where they identified which stakeholders were affected by that particular challenge and why, and which stakeholders may hold a solution to that challenge and why (Figure 1). The breakout groups' conclusions were subsequently fed back to and discussed by all workstream participants.

The challenges identified were grouped into five categories:

- Re-envisioning delivery of care in the future
- Who defines and designates hubs and spokes and how?
- Criteria for hubs and spokes
- Resource allocation
- Collaboration between hubs and spokes

1. RE-ENVISIONING DELIVERY OF CARE IN THE FUTURE

Precisely how the constitution and objectives of hub and spoke centres will be defined within Europe is likely to depend on local circumstances. The most familiar pattern is to have several regional hubs, each serving a number of local spoke centres. Alternatives include hubs that serve several small countries, a European network of hubs, and national or regional hubs that focus on advanced therapy medicinal products (ATMP), such as gene therapy, for all rare diseases or inherited and acquired bleeding disorders including haematological cancers.

Such versions of the hub and spoke model may transcend current regional and national arrangements and implementation would require political will and collaboration between multiple stakeholders. This has already been achieved in part with the introduction in 2017 of European Reference Networks (ERNs) [7]. These 24 virtual networks, which involve healthcare providers across Europe, aim to 'facilitate discussion on complex or rare diseases and conditions that require highly specialised treatment, and concentrated knowledge and resources'. The network for haematological disorders, EuroBloodNet, is developing a cross border referral system for patients with rare disorders, supported by standard management guidelines and education [8].

However, many existing networks cannot be incorporated into an ERN: ERNs can only include hospitals, so European-level networks such as EAHAD and European Haemophilia Safety Surveillance (EUHASS), which play a pivotal role in defining standards of care and advancing medical education in haemophilia and allied bleeding disorders, cannot be a part of EuroBloodNet. Adopting the ERN model may present a challenge to developed services, such as those for haemophilia, which are now relatively well resourced and have a strong infrastructure. While ERNs may help in building up networks for disease areas that do not have an existing European network, in the case of haemophilia they could slow down the process of redefining delivery of care through clashing with systems that are already in place.

Redesigning an existing bleeding disorders service, possibly within a larger network for the management of rare diseases, will involve technological change and collaboration across disciplinary boundaries. A multi-stakeholder approach will therefore be essential and should involve patients and their representative organisations, physicians, treatment centres and governments. There must be an effective flow of information to ensure effective delivery of care for patients regardless of whether they are attending a hub or spoke treatment centre. Here, key stakeholders will include IT providers and regulatory bodies in respect of information governance. Some patients, such as those with literacy issues, or language or cognitive difficulties, are difficult to reach in any model of care [9,10]. Their needs must be also considered in any new service configuration.

2. WHO DEFINES AND DESIGNATES HUBS AND SPOKES AND HOW?

There is currently little consensus about what constitutes an optimal hub and spoke model. Hubs may specialise in all rare bleeding disorders or specifically haemophilia, but they may also act as a centre of excellence in all rare diseases. The healthcare environments into which hub and spoke models will be introduced will vary between countries. Some countries already have strong established processes in terms of how centres for the treatment of bleeding disorders are certified, and the extent to which a hub and spoke model may be applied to existing services or whether it should replace them entirely remains unresolved. Consideration will need to be given at the European level regarding how far comprehensive care and the existing two-tiered accreditation system for treatment centres already resembles a hub and spoke model [11].

The hub and spoke design, the process of implementation and the stakeholders who contribute

to it will be influenced by current service levels. There is a risk that strategic decisions may be imposed by governments, regulators and payers who lack direct experience of rare diseases. Centralised strategic planning for a structured model must necessarily be carried out at a national level and involve patients, providers and payers. However, patient representative organisations and medical agencies should agree high-level criteria for service models that can be used in different countries to ensure parity of access to care and treatment. EAHAD, which defined the European Principles of Haemophilia Care [12], has a key role to play in providing these supranational definitions for hub and spoke designation. For smaller countries with smaller populations of people with bleeding disorders, it may be appropriate for this to involve a cross-border model, with spoke centres connected with a hub in a neighbouring country.

The scope of services to be provided also remains unresolved. Much of the discussion about reconfiguring services in haemophilia care has focused on gene therapy, but to what extent should this relatively small part of the care of people with bleeding disorders determine the objectives and activities of a hub and spoke model? Further, is it desirable – or feasible – for hubs to act as centres of expertise for all gene therapies? Relevant supranational agencies such as the European Medicines Agency should be involved in discussions about ensuring access to ATMPs.

3. CRITERIA FOR HUBS AND SPOKES

Linked to the definition and designation of hubs, and similarly related to ensuring parity of access to treatment and care across Europe, is the criteria for hubs and spokes.

The most obvious criteria for a hub and spoke service are the patient population and its geographical distribution. However, while these may determine the need for centres of expertise and their numbers, the patient population may change. We are accustomed to thinking about the prevalence of rare diseases as something that is constant, but this does not take into account the fact that, like all populations, people with rare diseases may change location. While the global or national prevalence may vary little, the number of people who make up a regional or local population with a rare disease need only change slightly to significantly affect the demand for a service. This is also an issue in countries with small populations and perhaps only a single treatment centre. Systems for data sharing between centres should be established to address this, and links should be established to enable referral to and connection with hub or reference centres in other countries as a means of bolstering local expertise. In developing service criteria, it is also essential to reconsider funding models based on population – this does not recognise excellence and therefore provides no incentive to achieve care quality standards.

These are challenges for governments and decision-making bodies, treatment centres and patient organisations. In aiming to ensure a uniform set of criteria that will underpin parity of access to treatment and care across Europe, the voices of both healthcare professionals and people with bleeding disorders must be heard. The EuroBloodNet ERN is well placed to help to develop the criteria which can inform funding assessments and the distribution of resources and would also provide a stronger platform for advocacy. The EUHANET initiative, a partnership of the EHC, EAHAD, University Medical College Utrecht, Medical Data Solutions and Services Ltd and the Fondazione IRCCS Ca' Granda in Milan, has developed a network of haemophilia centres that could provide a basis for moving forward [13,14].

4. RESOURCE ALLOCATION

Funding models will vary between countries but it is self-evident that hubs will require sufficient financial support to provide treatment with expensive medicines and for gene therapy. However, there is a risk that funding allocation will distort the way in which services as a whole develop. There is a concern that supranational recommendations on the number of centres of expertise in bleeding disorders in a given country may conflict with the existing numbers of centres funded by national governments. This could be used as a justification to withdraw funding, either at national level or because hospitals are reluctant to support a spoke centre that may not attract as much research funding. It could also impact centres' ability to attract investment from pharmaceutical companies for clinical research, and/or result in insurance companies favouring fewer centres based on their designation. Centres that are not recognised as centres of expertise in this way could therefore suffer multiple financial penalties. There is a risk that patient care may be compromised if support for smaller centres is cut.

The outlook is further complicated by the fact that centres are not all the same: they have expertise in different bleeding disorders – for example, one may

focus on haemophilia, another on von Willebrand disease. The designation of hub and spoke must take account of the nature and quality of the service.

One challenge in introducing a hub and spoke model is therefore how to avoid creating winners and losers. Some centres of expertise, such as those now providing gene therapy, will become a hub almost by default; others may seek hub status by developing their services. However, the scope for such development may be limited if hub status is defined by population-based criteria. Potential hubs may choose to build partnerships with centres likely to become spokes or to prioritise their own development and retain control over funding. Centres potentially facing designation as a spoke may face disinvestment and therefore be reluctant to participate in a process that could lower their status and capability.

The impact of introducing a hub and spoke model will depend on how well developed the bleeding disorders service is. In The Netherlands, for example, there are currently eight centres providing a high-level service but fewer may be funded if external service criteria are applied.

This poses challenges in developing the best criteria for a bleeding disorders service, agreeing which agencies have the power to impose change, and pitting centres against one another in a competition for resources. Centres will require sufficient resources to provide high quality care for their populations without, in effect, shifting costs onto patients by making them travel further and spend more time accessing care. Any redistribution of financial and human resources must be undertaken in a balanced manner. Greater use of new technologies could be one option for maintaining services [15-17]. The key stakeholders in these decisions will be patients' and doctors' organisations, governments, treatment centres, health insurance agencies and the pharmaceutical industry.

5. COLLABORATION BETWEEN HUBS AND SPOKES

There will be a variety of ways in which hubs and spokes work together. For example, there are likely to be few hubs for gene therapy and they may provide treatment for patients with any rare disease, not only bleeding disorders. Spokes that primarily manage bleeding disorders will therefore be involved in only part of the hub's activities. Further, a hub may not provide all aspects of a gene therapy service – for example, where expertise has been developed locally, a spoke may become a more equal partner in sharing service delivery.

The hub and spoke model required for gene therapy is likely to be more complex than the arrangements currently in place or that may be needed for bleeding disorders [18,19]. The high cost, resources and clinical expertise necessary for gene therapy mean that the work of a hub is fundamentally different from that of a spoke. By contrast, the activities of comprehensive treatment centres are similar in nature to those of local treatment centres. Further, some comprehensive treatment centres have developed additional expertise in related services such as thrombosis management that would enhance their role as a hub.

These differences emphasise the importance of designing a hub and spoke model from the patient's perspective, taking into account the individual's journey along the care pathway. A person undergoing gene therapy may have multiple consultations at different sites, and this process will be different from their experience of receiving care for a bleeding disorder at one location. There should be effective signposting within a hub and spoke model and clearly defined roles for clinicians. This will require efficient communication between hubs and spokes, and should ensure that patients do not receive confusing or conflicting messages and are not overloaded with excessive information.

These challenges should be addressed by patients, caregivers, clinicians, treatment centres and regulators. Patient organisations should ensure that patients are provided with information that empowers them to contribute to decision-making throughout their care pathway. In turn, hubs and spokes will need to develop a common approach, perhaps facilitated by EAHAD [18], and optimise their use of information technology, drawing on the experience of central government in data management where possible.

SUMMARY AND NEXT STEPS

Hub and spoke models of care for rare diseases can deliver high quality care for relatively low numbers of people over a wide geographical area. Some examples are already established but further development should be informed by defining the criteria for hub and spoke status, prioritising patients in service reorganisation and in the care pathway, and considering the impact of introducing a different service model into a developed health service compared with establishing a new service. The next step is to vet the challenges identified by this workstream with a broader group of external stakeholders and bring their perspectives back for consideration.

THE EHC THINK TANK

The European Haemophilia Consortium (EHC) Think Tank was launched in June 2021. Building on existing advocacy activities, the initiative brings together a broad group of stakeholders who will engage with key thematic areas or workstreams identified as priority areas for "systems change" within European healthcare systems [17]. The EHC Think Tanks seeks to mobilise the agency and purpose of all stakeholders in the healthcare system to collectively design and champion potential solutions to existing problems.

The EHC steering committee was presented with more than 20 topic areas identified from patient, medical and scientific volunteers within the broad community. Following a prioritisation process in early 2021, three key topic areas were identified for Think Tank workstreams to tackle:

- Registries
- Hub-and-spoke treatment models
- Patient agency.

Workstream members are invited based on their expertise and potential for constructive engagement, including patient and industry perspectives alongside a balance of healthcare professional, academic, regulatory, governmental and geographical representation. All workstream activities are held under the Chatham House rule to enable inclusive and open discussion: participants are free to use the information received, but neither the identity nor the affiliation of the speakers, nor that of any other participant, may be revealed [18].

Each is project-managed from within its individual membership. Members will set their own agendas, timelines, and targeted outputs, with operational, logistical, methodological and facilitation support from EHC staff and Think Tank practitioners.

While concrete outcomes and results will vary across workstreams, they are likely to include (but not be limited to) manuscripts, consensus-based guidelines, monographs, white papers, and so on.

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ORCID

Amanda Bok *https://orcid.org/0000-0001-7303-8833*
Declan Noone *https://orcid.org/0000-0003-2183-4277*
Naja Skouw-Rasmussen *https://orcid.org/0000-0002-7845-6230*

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