Canadian Integrated and Comprehensive Care Standards for Inherited Bleeding Disorders

[Draft 4 – January 15, 2020]

Authored by the Canadian Inherited Bleeding Disorder Standards Working Group

Endorsed by

Canadian Hemophilia Society
Help Stop the Bleeding

AHCDC

Canadian Association of Nurses in Hemophilia Care

Canadian Social Workers in Hemophilia Care

CANADIAN PHYSIOTHERAPISTS IN HEMOPHILIA CARE

PHYSIOTHÉRAPEUTES CANADIENS EN HÉMOPHILIE

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INTRODUCTION

The *Integrated Care* model, also known as the *Comprehensive Care* model or the *Hemophilia Treatment Centre (HTC)* model

"This term refers to the continuous supervision of all medical and psychosocial aspects affecting the haemophilia patient and family, including supervision of home-based treatment (either prophylaxis or on-demand). It generally demands that all modalities of care – access to care providers, as well as diagnostic and therapeutic facilities – are available and delivered in a single specialized centre, by a team of health care providers. The composition of the integrated care team varies, but generally adheres to key components recommended by the World Federation of Haemophilia, including a medical director, a nurse coordinator, a physical therapist, a psychosocial expert (e.g. social worker) and a specialized coagulation laboratory."
- NHF-McMaster Guideline on Care Models for Haemophilia Management[1]

The comprehensive management of hemophilia began as "the Hemophilia Demonstration Project" at the Los Angeles Orthopedic Hospital in 1964. "*The project represented a 'first' in hemophilic rehabilitation since it combined in one coordinated setting all the professional specialists required to diagnose and to treat the multiple problems manifested by this disease."*[2]

In Canada, the comprehensive care model was first promulgated in the conference entitled "Comprehensive Care for the Canadian Hemophiliac", held in Winnipeg in May 1978, organized by the Canadian Hemophilia Society (CHS) in close collaboration with its own multidisciplinary Medical and Scientific Advisory Committee (MSAC).[3]

It has become the accepted model of hemophilia care throughout the world and is recommended by the World Federation of Hemophilia in its *Guidelines for the Management of Hemophilia*, 2nd Edition.[4] [5]

[N.B. The balance of this document will be inclusive and refer to the entire family of inherited bleeding disorders rather than exclusively to hemophilia, to people with bleeding disorders [PwBD] and to treatment centres rather than Hemophilia Treatment Centres.]

The First Edition of the *Canadian Comprehensive Care Standards for Hemophilia and Other Inherited Bleeding Disorders* was published in June 2007. It has proven invaluable in guiding health care providers in treatment centres, hospital administrators, ministries of health and the patient association in the coordination and delivery of "integrated" or "comprehensive" care for PwBD.

Much, however, has changed since then, notably:

- a doubling in the number of diagnosed patients over 15 years to more than 10,000, largely through increased recognition of abnormal bleeding symptoms and improved diagnosis[6]
- an increase in co-morbidities due to an ageing patient population, necessitating enhanced collaboration with specialty services such as cardiology, oncology and vascular surgery
- the number and complexity of treatment regimens (e.g. immune tolerance induction to eradicate inhibitors, extended half-life factor concentrates, non-factor replacement therapies …)
- the introduction of new clinical assessment tools such as point-of-care ultrasound and standardized hemophilia-specific outcome measures
- the development and adoption of improved tools to monitor and manage care, such as comprehensive registries, expanded pharmacokinetic capabilities, electronic patient files and tele-medicine

- an emphasis on increased participation in physical activities, physical therapy and orthopedic interventions
- increased health system requirements for accountability for utilization of health resources (e.g., treatments, human resources, laboratory testing)
- an increasing obligation to collect real-world evidence on health outcomes and an expectation that treatment centres participate in registries and research
- an increased recognition of the impact of bleeding disorders in women, resulting in increased diagnosis, and necessitating improved collaboration with specialties such as obstetrics and anesthesia to coordinate care plans
- an increased recognition of the complexity and impact of mild bleeding disorders
- an increased recognition of the level of pain experienced by many PwBD and the need to manage it
- an increased recognition of the psychosocial issues for both patients and caregivers related to being diagnosed and living with a chronic condition
- the introduction in Canada of frequent product tendering and wholesale treatment product switching for patients, imposing on treatment centres the obligation to review, consent and implement new treatments in hundreds of patients every two to three years
- an increasing population of new Canadians with bleeding disorders who arrive with joint damage and whose care requires cultural and linguistic sensitivity.

An updated version, reflecting current best practices, is needed.

The intended audience for these standards is:

- treatment centre team members
- health care administrators including:
    - department and program managers
    - institutional executives
    - regional and provincial health authorities
    - ministries of health
- PwBD and caregivers
- researchers
- Canadian Blood Services and Héma-Québec
- the Provincial/Territorial Blood Liaison Committee.

METHODOLOGY

Multidisciplinary input

This version of the standards was written by the Canadian Inherited Bleeding Disorder Standards Working Group (CIBDSWG). It includes:

- Association of Hemophilia Clinic Directors of Canada
  - Dr. Lawrence Jardine, (co-chair), Ontario
  - Dr. Natasha Pardy, Newfoundland and Labrador
- Canadian Association of Nurses in Hemophilia Care
  - Karen Sims, NP(A), British Columbia
  - Marie-Hélène Thompson, B. Sc. N, Québec
- Canadian Hemophilia Society
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  - Wendy Quinn, NP, Saskatchewan
  - Pamela Wilton, RN, Ontario
- Canadian Physiotherapists in Hemophilia Care
  - Laurence Boma-Fisher, PT, Ontario
  - JoAnn Nilson, PT, Saskatchewan
- Canadian Social Workers in Hemophilia Care
  - Jennifer King, SW, Saskatchewan
- Ontario Regional Blood Coordinating Network
  - Sarah Crymble, Hemophilia Provincial Coordinator, Ontario

These representatives consulted widely with their respective organizations.

International standards

The CIBDSWG reviewed standards and guidelines produced by the World Federation of Hemophilia, the UK Haemophilia Alliance, the European Haemophilia Network and the U.S. National Hemophilia Foundation, (see Appendix 1, International standards for the care and treatment of inherited bleeding disorders) as well as Canadian standards from other conditions, for example cystic fibrosis.

Literature review

Literature searches were conducted to identify research support for the standards proposed.

External review

External reviews were invited from Dr. Paul Giangrande (UK) and Dr. Carol Kasper (USA).

GOALS

These standards are designed to:

- ensure all Canadians with an inherited bleeding disorder, regardless of geographical location, are registered with treatment centres providing evidence-based and standardized care.
- ensure delivery of team-based integrated care.
- provide a measurement tool for delivery of care and inform ongoing advances in patient care in order to …
    - allow benchmarking across quality assurance indicators;
    - promote consistent measurement of health outcomes;
    - reduce health-related risks such as joint disease, inhibitors and treatment-related complications that may result from non-integrated or outdated practices, through the comprehensive care model and open collaboration between treatment centres, hospitals, health authorities and governments.
- provide guidance to those responsible for coordinating the delivery of care to PwBD.
- ensure that local frontline health care providers can access and consult with experienced health care providers in those treatment centres.
- empower PwBD and caregivers to be engaged in their care and assist treatment centre personnel to equitably and consistently meet their expectations.
- ensure that health resources are used efficiently and effectively.
- promote value-based health care.

POPULATIONS SERVED

This document applies to bleeding disorder treatment centres who serve people, caregivers and families living with …

- hemophilia A and B, including carriers;
- inhibitors to clotting factors, including acquired coagulopathies;
- von Willebrand disease;
- rare factor deficiencies;
- inherited platelet function disorders.

N.B. Treatment centres may also serve other congenital disorders associated with a bleeding tendency (e.g. Ehlers-Danlos Syndrome, Hereditary Hemorrhagic Telangiectasia …).

If a treatment centre is expanded because of synergies to serve other conditions, for example, hemoglobinopathies, hereditary angioedema, primary immune deficiency, etc., additional resources will be required; these standards do not apply to these conditions.

PRINCIPLES OF COMPREHENSIVE INTEGRATED CARE

These standards are based on the following principles:

- Treatment centres shall strive to register and oversee care to all PwBD living in the treatment centre's geographical region.
- Treatment centres shall respond to the complexity and rarity of inherited bleeding disorders, and the associated lifelong needs by delivering care through an integrated, multi- and inter-disciplinary chronic disease management model.
    - Treatment centres shall monitor for complications, including joint disease, psychosocial issues, inhibitors, adverse drug reactions and transfusion reactions.
    - Health promotion and disease prevention in the context of bleeding and its consequences shall be a priority.
    - Care shall be delivered in a way that aims to minimize the pain, incapacity, and physical and emotional disability that may result from inherited bleeding disorders and their complications.
    - A holistic approach to patient care shall be adopted, ensuring that all PwBD have access to a wide range of specialist services.
- Treatment centres will provide care that is based on the best available evidence.
- Services shall be individualized and delivered such that PwBD and families are partners in care.
    - The risks and benefits of all treatment options shall be presented and discussed.
    - Shared decision-making shall be promoted.
    - Home-based care shall be facilitated, where appropriate.
    - Care shall be delivered/provided within the person's own community, wherever achievable in a safe and suitable manner.
- Health care providers shall behave in a manner that is culturally and socially respectful.
    - Privacy and confidentiality shall be respected.
- Treatment centres shall participate regularly in quality assurance activities (e.g. contributing data to a national report with provincial/treatment centre breakdown that is then sent to program managers, health authorities, governments and patient associations).

STANDARDS

A. Role of treatment centres

The treatment centre shall …

1. be supported by the host institution and health authority to achieve the obligations set out in this document, and together with health administration maintain/pursue support and recognition by the provincial Ministry of Health.
2. be affiliated with an academic health care institution and engage in relevant continuing education opportunities.
3. raise awareness in the local medical community to facilitate referrals and collaborate in care.
4. strive to register and provide care to all PwBD and their families in its respective region.
5. provide access to the full range of expertise required to respond to the complex and diverse needs of PwBD. (See Appendix 2, Team members and roles: Core and extended.)

B. Diagnosis

The treatment centre shall…

1. have a triage process for new patient referrals.
2. have access to an accredited hemostasis laboratory with appropriate arrangements in place for 24/7 coagulation testing and establish formal links for specialized hemostasis testing when required. (See Appendix 6, Coagulation testing.)
3. have access to appropriate transfusion medicine and blood bank services.
4. have access to a diagnostic imaging department and establish formal links where specialized radiology services are required.
5. have a formal relationship with a genetic laboratory so that all patients and families have access to carrier testing and genetic testing.

C. Care

The treatment centre shall …

1. provide access to the full range of services required to respond to the complex and diverse needs of PwBD.
2. provide care and treatment based on the most current clinical treatment guidelines.[5 7] Where guidelines are not available, care shall be based on the most current literature and best practices.
3. develop and maintain written policies and procedures for the treatment of non-urgent, urgent and emergency bleeding episodes.
4. make prophylaxis and home therapy available to PwBD as appropriate.
5. provide 24/7 access to bleeding disorder expertise to PwBD through the centre, through partnerships, on call or consultative services.
6. inform PwBD and their caregivers whom to contact in the event of a bleeding episode.
7. coordinate treatment and follow-up for acute bleeds.
8. offer regular comprehensive clinical assessments for all PwBD regardless of type of bleeding disorder or severity. (See Appendix 5, Recommended schedule for assessments.)

9. collect detailed health outcomes over time. (See Appendix 4, Health outcomes to be measured.)
10. maintain a Patient Education Record for all PwBD.
11. have procedures to ensure that care is coordinated during transition (pediatric to adult, program to program, or province to province) to ensure continuity of care.
12. have access to staff with sufficient clinical skills to deal with all PwBD in a manner that is appropriate to their age and condition.
13. provide psychosocial support of the PwBD and family regarding schooling, employment and relationships, and the provision of social supports and counselling services.
14. provide advice and support to ageing PwBD who require treatment for co-morbidities and to the health care providers who care for them.
15. provide access to appropriate hematologic, obstetric and gynaecologic services to women with inherited bleeding disorders, including carriers of hemophilia.
16. provide information to PwBD and their families about clinical phenotype, therapeutic options and genetic information relevant to their diagnosis.
17. provide genetic counselling delivered by an individual trained in genetic counselling and knowledgeable about inherited bleeding disorders.
18. offer genetic testing to confirm the status of potential carriers of inherited bleeding disorders. Tests shall be available to all potentially affected female members of families with inherited bleeding disorders.
19. provide up-to-date documentation (e.g. *FactorFirst* wallet card) that identifies each person's bleeding disorder and recommended treatment plan.
20. provide to PwBD/caregivers comprehensive information on treatment benefits and risks to make informed decisions.
21. offer educational information on current issues related to bleeding disorders to PwBD and their families.
22. provide information to PwBD and their families about clinical phenotype, therapeutic options and genetic information relevant to their diagnosis. For PwBD/families on home therapy, the centre shall provide comprehensive training about safe and responsible handling, storage, preparation and administration of treatment products, safe disposal of used equipment and supplies.
23. ensure that PwBD have access to infusion/injection equipment and supplies, either by supplying or facilitating procurement.
24. respect all professional, institutional and provincial guidelines regarding patient privacy and confidentiality.

D. Product monitoring

The treatment centre shall…

1. maintain a list of all PwBD registered in the home therapy program.
2. ensure that PwBD on home therapy have access to the required supply of factor concentrates and non-factor replacement therapies.
3. establish formal links to Canadian Blood Services or Héma-Québec for product ordering.
4. be able to…
    a. track home inventory in real time;
    b. enter home inventory in electronic reporting systems;

c. account for and report the use of all factor concentrates and non-factor replacement therapies distributed to their registered patients;
d. monitor expiry dates of all factor concentrates and non-factor replacement therapies distributed to their registered patients;
e. be able to recall products within 24 hours.

5. establish a mechanism whereby PwBD and their families record all bleeding episodes and treatments with clotting factor concentrates and non-factor replacement therapies so as to provide the treatment centre essential clinical data. As a condition of receiving home therapy, PwBD and their families shall agree to provide this information.
6. report adverse events that occur in association with the administration of coagulation factor concentrates or non-factor replacement therapies, as mandated by the regulator, manufacturer or other appropriate body.

E. Human resources for integrated care

The treatment centre shall…

1. maintain a complete and adequate complement of core team members. (See Appendix 2, Team members and roles: Core and extended.) The core team members shall have the appropriate training and qualifications to provide care to the patient population. (See Appendix 7, Standards of care for core disciplines in bleeding disorder care.)
2. have a process to monitor its ability to deliver care to the patient population and to request adjustments in human resources, when necessary.
3. provide a mechanism for team members to share knowledge with each other to promote best patient outcomes.
4. develop and maintain a referral list for extended team members and utilize their services as needed.
5. invite extended team members to organize educational workshops and activities as appropriate.
6. demonstrate collaboration among all team members.
7. facilitate core team members being members of relevant organizations and / or working groups within the bleeding disorder community.
8. facilitate access to reference materials for team members and students (e.g. *AHCDC Clinical Practice Guidelines*, journal articles and texts).

F. Physical resources

The treatment centre shall…

1. work with the hospital administration to ensure a dedicated clinical area that is age-appropriate, comfortable, quiet and adequately equipped, that respects privacy and confidentiality, and that is designed for people with disabilities or mobility aids.
2. be located in a facility that is linked with a day medicine department and an emergency department so that PwBD can obtain treatment 24/7.

G. Information systems, health records, data collection

The treatment centre shall…

1. follow provincial and institutional policies for maintaining records and assign a confidential identifier to each patient.
2. document each patient's treatment plan, as appropriate, and update it annually.
3. be registered with a bleeding disorder registry database (e.g. CBDR, iCHIP) and ensure that all core team members have access to the information systems.
4. keep data current and routinely export data, as required, to the provincial and national databases.
5. collect detailed information concerning the outcomes of treatment. (See Appendix 4, Health outcomes to be measured.)

H. Linkages

The treatment centre shall…

1. provide information on inherited bleeding disorders, contact information in case of emergency and treatment recommendations to Emergency Departments and family physicians.
2. provide contact information and emergency treatment recommendations to the Emergency Department nearest to the PwBD's home.
3. provide education and treatment recommendations to other community professionals who provide services to PwBD (e.g. surgery, dentistry).
4. establish a process for referring PwBD to services not provided within the program.
5. have a process in place to meet and discuss issues of mutual concern with the Canadian Hemophilia Society and its provincial chapters.
6. maintain current contact information for the treatment centre in listings with the Canadian Hemophilia Society, the World Federation of Hemophilia and parent hospital.

I. Accreditation, audit, quality assurance

The treatment centre shall…

1. participate in hospital or peer evaluation and respond to critical appraisal.
2. participate in a formal accreditation and evaluation process, when established.
3. make known the institution's process to allow PwBD and families to communicate concerns, complaints and appreciation.
4. actively advocate for adequate physical and human resources to meet these standards.

APPENDIX 1

International standards for the care and treatment of inherited bleeding disorders

- The U.K. Haemophilia Alliance: A National Service Specification for Haemophilia and Other Inherited Bleeding Disorders, 2006
- U.K. NHS Standard Contract for Haemophilia, 2013/14
- The European Standards of Haemophilia Centres, Giangrande et al. Blood Transfus 2014; 12 Suppl 3: s525-30 DOI 10.2450/2014.0056-14s
- World Federation of Hemophilia Guidelines for the Management of Hemophilia, 2nd Edition, 2012
- U.S. National Hemophilia Foundation Standards and Criteria for the Care of Persons with Congenital Bleeding Disorders, 2002
- UK Standards of Care Service Provision of Physiotherapy for Adults with Haemophilia & other Inherited Bleeding Disorders, 2017
- UK Standards of Care Service Provision of Physiotherapy for Children with Haemophilia & other Inherited Bleeding Disorders, 2017

APPENDIX 2
Team members and roles: Core and extended

Core team

In order to provide integrated care, the following disciplines are required in each treatment centre:

Medical director (an adult or pediatric hematologist or internist)
Provides subspecialty bleeding disorder medical expertise, supervises medical care and takes leadership in the treatment centre.

Nurse (RN or NP)
Provides expert clinical nursing care for PwBD, practices/acts within the domains of education, administration, research and policy to improve care delivery, may be the primary point of entry to facilitate access to care.

Physiotherapist
Monitors growth and development, offers a range of assessments and management for PwBD with acute and chronic conditions resulting from musculoskeletal bleeds, is responsible for monitoring joint function on a long-term basis, addresses prevention of bleeds and joint damage, promotes physical activity and coordinates rehabilitation.

Social worker
*P*rovides support through psychosocial assessments, counselling/psychotherapy, advocacy, patient, family and community education, resources, supports and discharge planning.

Coagulation laboratory staff
Perform diagnostic testing, and accurate and precise coagulation assays and inhibitor testing.

Blood bank staff
Prepare, store and distribute coagulation products.

Administrative assistant / data manager
Supports the proper functioning of the treatment centre and collates clinical and financial data.

Extended team

In order to provide integrated care, the following disciplines should be available when they are needed for individual patients:

Primary care provider
Provides primary health care.

Orthopedic surgeon
Monitors joint and muscle problems and identifies the need for surgical intervention.

Rheumatologist
Provides diagnosis (detection) and treatment of musculoskeletal disease.

Hepatologist or gastroenterologist
Monitors and treats the complications from HCV infection.

Infectious disease specialist
Monitors and treats the complications of transmissible and blood-borne infections such as HIV.

Gynaecologist/Obstetrician
Manages the care for women with bleeding disorders: antenatal care for carriers, the delivery of a child who has a bleeding disorder, and other gynaecological issues.

Geneticist
Coordinates genetic testing and mutational analysis to enable confirmation of diagnosis, determination of carrier status and antenatal fetal testing.

Genetic Counsellor
Supports PwBD and families regarding transmission, conception and family planning.

Dentist
Provides specialized dental services and advises community dentists.

Expert in pain management
Supports PwBD who need specialized pain management programs.

Psychiatrist/ Psychologist
Provides specialized psychosocial support and treatment.

Child-life specialist
Supports families in dealing with the challenges of a chronic disease in a young child.

Athletic therapist/kinesiologist/Sports medicine specialist/Physical medicine specialist
Guide the PwBD /caregiver in activities that safely promote fitness.

Geriatrician
Provides expertise and care in health challenges due to ageing.

Occupational therapist
Promotes independence, meaningful occupations, and the PwBD's functional ability to achieve daily routines/roles of choice.

APPENDIX 3
Staffing levels: Complexity matrix

The authors of the standards consider that basing staffing levels solely on numbers of patients (e.g. numbers of people with severe hemophilia A and B) is overly simplistic and not representative of the complexity of treating PwBD in Canada with its geographical, cultural and linguistic challenges. We recommend that the following factors be taken into account when establishing optimal staffing levels.

1. Geography – In provinces with one designated treatment centre, or where PwBD may live a considerable distance from the centre, there will be a need for outreach clinics and the availability of Telehealth.
2. Number of PwBD
3. Case mix of PwBD:
    a. Proportion of severe vs. mild bleeding disorders
    b. Number of rare bleeding disorders
    c. The proportion of males vs. females
    d. The proportion of pediatric vs. adults vs. seniors: availability of appropriate facilities/expertise available for all
        i. Need for transition care
    e. The number of newly diagnosed
    f. The number on home infusion programs
    g. The number with inhibitors
    h. The number with co-morbidities including arthropathy, HIV, HCV infection, ageing
    i. The number with chronic pain issues
    j. The number requiring orthopedic surgery (coordinating external consultations, infusions, coagulation testing, pain management, rehabilitation, home care …)
    k. The proportion of new Canadians: language and cultural issues, pre-existing multiple arthropathies
    l. The number with venous access issues, central venous access devices
    m. Education levels, literacy levels and active participation in their own care
4. Other:
    a. Changes in treatment products
    b. Social determinants of health
    c. Centre involvement in research

APPENDIX 4
Health outcomes to be measured

The treatment centre shall collect the following data (as a minimum):

- Date of clinic review
- Date of updated treatment plan
- Total units of clotting factor concentrate used by each patient per year
- Total number of treatments received by each patient per year
- Amount of coagulation factor concentrate given for prophylaxis, on demand, surgery, ITI
- Pharmacokinetic testing to establish recovery, half-life and trough levels in prophylaxis
- Total number of annual bleeds
- Total number of inpatient days due to bleeds
- Inhibitor status
- HIV/HCV status
- Clinical outcomes of surgical and dental procedures performed, including bleeding outcomes and units of coagulation factor concentrates used
- Musculoskeletal assessments, including measures of impairment, function and participation, and number of target joints (reference: WHO International Classification of Functioning)[8]
- Psychosocial assessments including supports, family history, gaps in care, career status, current mental health status and impact of diagnosis on daily living

APPENDIX 5
Recommended schedule for assessments

To come

APPENDIX 6
Coagulation testing

List of coagulation tests that should be accessible by treatment centres and their anticipated turn-around-times:

LAB TEST	Locally accessible	24-hour service	TAT	Locally accessible or accessible through link with specialized laboratory
CBC	Yes	Yes	Within 1 hour	
ABO blood type	Yes	Yes	Within 1 hour	
PT/INR, aPTT, thrombin time, fibrinogen	Yes	Yes	Within 1 hour	
Mixing studies	Yes		Within 3 hours	
Factor VIII and IX one-stage assays	Yes	Yes	Within 3 hours	
Factor VIII and IX chromogenic assays	*[1]			Yes
Inhibitor assay	Yes		Within 12 hours	
Anti-porcine FVIII inhibitor assay				Yes
VWF antigen and activity assay	Yes		Within 3 hours	
VWF Multimers				Yes
Collagen binding activity assay; VWF-FVIII binding assay; VWFpp plasma concentration (VWFpp/VWF:Ag)				Yes
Factor II, V, VII, X, XI and XIII activity assays	Yes		Within 6-12 hours	
Inhibitor assays for rare coagulation factor deficiencies				Yes
Platelet aggregation, RIPA				Yes
PTEM and platelet flow cytometry				Yes

* Where local patients are using extended half-life products and FVIII mimetics, we encourage the local lab to invest in required chromogenic equipment and to develop the necessary experience to appropriately monitor patients locally.

APPENDIX 7

Standards of care for core disciplines in bleeding disorder care

To come.

References

[1] NHF-McMaster Guideline on Care Models for Haemophilia Management. Pai M et l. Haemophilia (2016), 22 (Suppl. 3), 6–16

[2] Comprehensive Management of Hemophilia, Donna Boone editor, FA Davis Company, 1976. ISBN 0-8036-1000-9

[3] The haemophilia certification system in Canada, D Matino et al, Blood Transfus 2014; 12 Suppl 3: s531-41 DOI 10.2450/2014.0032-14s

[4] Guidelines for the management of hemophilia, A Srivastava et al, Haemophilia (2012), 1–47 DOI: 10.1111/j.1365-2516.2012.02909.x

[5] Developing models of haemophilia care. A Street, Haemophilia (2012), 18 (Suppl. 4), 89–93 DOI: 10.1111/j.1365-2516.2012.02876.x

[6] Canadian Hemophilia Registry, Data summaries, www.fhs.mcmaster.ca/chr

[7] National benchmarking of paediatric haemophilia treatment centres, Research and Practice in Thrombosis and Haemostasis. Conference: 27th Congress of the International Society on Thrombosis and Haemostasis. Australia. 3 (Supplement 1) (pp 890), 2019. Date of Publication: July 2019

[8] Choosing outcome assessment tools in haemophilia care and research: a multidisciplinary perspective, K Fischer et al, Haemophilia (2017), 23, 11–24 DOI: 10.1111/hae.13088