Range of Motion Data Collected in the ATHNdataset: Completeness, Variability, and Temporality

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Introduction

- Limited literature exists on the long-term trajectory of joint health and functionality experienced by people with hemophilia A
- Range of motion (ROM) is a measure of joint functionality that was historically collected during the Centers for Disease Control and Prevention's Universal Data Collection (UDC) program from May 1998 through 2011; these data are recorded in the ATHNdataset
  - ROM data in the ATHNdataset are a potentially unique source of joint function data, as ROM data collection continued after 2011 at some Hemophilia Treatment Centers
- Here, we assessed the quality of joint ROM data in the ATHNdataset to understand the feasibility of describing longitudinal changes in ROM among the overall population and in various subpopulations of people with hemophilia A

Methods

- The ATHNdataset is a HIPPA-compliant, de-identified patient health dataset containing data from individuals with bleeding and clotting disorders receiving care through ATHN affiliates.[1] We queried the ATHNdataset to obtain joint ROM data for people with hemophilia A
- Data completeness was described in an overall population that included adult male individuals with hemophilia A currently consented for the ATHNdataset who had ≥3 ROM assessments recorded between 5/1/1998 and 12/31/2019, with ≥2 years between first and last ROM assessments
  - By adjusting the inclusion/exclusion criteria, additional subpopulations were explored to address observed missingness and potential confounders
- A ROM assessment was considered complete if values were recorded for right and left measurements of ankle plantarflexion and dorsiflexion; elbow extension, flexion, pronation, and supination; hip extension and flexion; knee extension and flexion; and shoulder flexion
- Potential sources of missing data related to temporal trends, reported reasons of missingness, and disease severity were explored

Results

- Overall, 1989 people with hemophilia A who met the inclusion criteria were identified in the analysis population in the ATHNdataset **(Table 1)**
  - Additional subpopulations were considered with the aim of minimizing the amount of missing data

Table 1. Cohorts used in the analysis and rationale

Cohort definition	Cohort rationale	Sample size
Full cohort for inclusion criteria	Original protocol cohort	1989
Subpopulations including participants with		
■ Complete ROM assessments at first and last visits	Minimizing missingness & maximizing longitudinally	189
■ ≥3 visits with complete ROM assessments ■ First and last visits ≥2 years apart ■ Age >18 years at last visit	Original cohort minimizing missingness	381
■ ≥3 visits with complete ROM assessments	Minimizing missingness (allowing all ages) in >2 ROM assessments	412
■ Complete ROM assessments at first and last visits ≥2 years apart ■ Age >18 years at the last visit	Minimizing missingness in ≥2 ROM assessments (allowing all ages, providing ≥18 by last measure)	496
■ Complete ROM assessments at first and last visits ≥2 years apart	Minimizing missingness in ≥2 measures (allowing all ages)	508
■ ≥2 visits with complete ROM assessments (without considering REE and RKE) ■ Age >18 years at the last visit ■ ≥2 years between complete visits	Original cohort ensuring complete measures (except for measures with systematic missingness) at ≥2 ROM assessments	659
■ ≥1 visit with complete ROM assessment	Minimizing missingness in ≥1 ROM assessment for potential intra-person extrapolation	1072
■ ≥1 measure at each anatomical location	Minimizing missingness in ≥1 ROM assessment for potential intra-person extrapolation (assuming no differences in anatomical location between sides)	1311

Complete ROM visits included recorded flexion/plantarflexion and extension/dorsiflexion measurements on both sides for the ankle, knee, hip, and elbow, as well as shoulder flexion on both sides. REE, right elbow extension; RKE, right knee extension; ROM, range of motion.

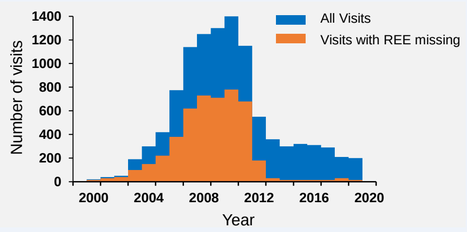
Table 2. Percent of participants with complete measures by joint and side at first and last ROM assessments

	Hemophilia A severity							
	Overall		Severe		Moderate		Mild	
Assessment	First	Last	First	Last	First	Last	First	Last
L ankle dorsiflexion	91.3%	90.2%	90.2%	89.1%	91.6%	90.0%	95.6%	95.0%
R ankle dorsiflexion	84.8%	86.8%	82.7%	85.0%	85.1%	87.7%	93.4%	93.4%
L ankle plantarflexion	97.3%	92.9%	97.1%	92.4%	96.8%	92.6%	98.7%	95.3%
R ankle plantarflexion	95.9%	92.1%	95.6%	91.3%	95.1%	92.9%	98.1%	94.3%
L elbow extension	57.5%	76.2%	63.4%	81.5%	46.6%	65.7%	42.6%	63.7%
R elbow extension	44.5%	68.9%	48.8%	73.8%	36.6%	57.6%	33.8%	58.7%
L elbow flexion	98.6%	96.0%	98.4%	95.7%	98.7%	96.1%	99.4%	97.2%
R elbow flexion	97.7%	95.4%	97.3%	95.0%	98.1%	95.5%	99.4%	96.8%
L elbow pronation	97.6%	88.2%	97.4%	87.3%	97.4%	89.0%	98.7%	91.2%
R elbow pronation	96.6%	87.4%	96.2%	86.4%	96.8%	88.3%	98.4%	90.9%
L elbow supination	97.2%	89.0%	96.9%	87.7%	96.8%	90.0%	98.7%	93.7%
R elbow supination	95.5%	88.1%	94.6%	86.6%	96.1%	89.6%	98.4%	93.1%
L hip extension	94.2%	83.8%	94.0%	80.6%	94.2%	88.3%	95.0%	93.1%
R hip extension	92.8%	83.1%	92.9%	80.1%	91.9%	86.4%	93.4%	92.4%
L hip flexion	97.1%	88.0%	96.6%	85.7%	98.1%	91.6%	98.7%	94.3%
R hip flexion	96.6%	87.9%	96.4%	85.7%	97.4%	91.6%	96.5%	93.7%
L knee extension	41.2%	67.7%	44.2%	71.0%	33.3%	59.9%	35.6%	61.2%
R knee extension	31.0%	60.3%	32.6%	62.9%	25.9%	54.0%	29.3%	55.5%
L knee flexion	98.0%	95.2%	97.9%	94.5%	98.4%	96.1%	98.1%	97.2%
R knee flexion	96.6%	94.6%	96.2%	93.8%	97.7%	96.1%	97.5%	96.5%
L shoulder flexion	99.2%	94.2%	99.1%	93.0%	99.0%	96.4%	99.7%	97.2%
R shoulder flexion	98.7%	94.3%	98.8%	93.0%	98.1%	96.8%	98.7%	97.2%

Data are for the full cohort (n = 1989). Darker values indicate higher percentages of participants with missing measures. L, left; R, right; ROM, range of motion.

- In many visits where ROM was assessed, at least 1 measure was missing. Missingness was particularly high for extension values in the knees and elbows, with differing proportions of missingness between the right and left sides of the body (**Table 2**)
  - When evaluating just the first and last available ROM assessments, up to 70% of individuals had missing values across joints/sides
  - Missingness did not appear to differ by disease severity
- The high proportion of missingness for extension values in particular joints suggested that missingness was systematic rather than random; thus, potential causes for missing data were further investigated
- Missing data did not appear to be due to the transfer of data between data entry systems comprising the ATHNdataset, nor due to changes in funding and training after UDC, as missingness was generally proportional to the number of assessments performed over time (**Figure 1**)

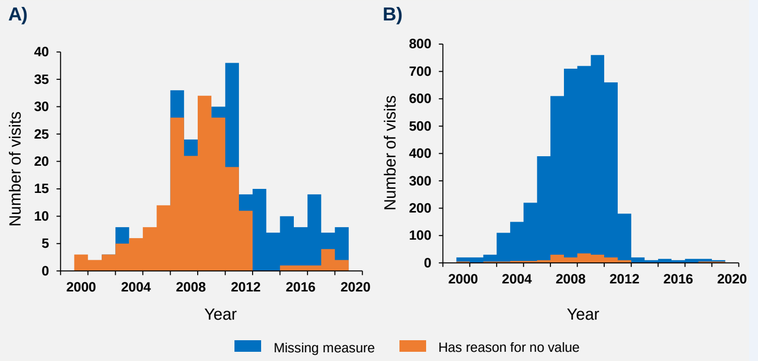
Figure 1. Missing right elbow extension measures over time



Data are for the full cohort (n = 1989), including both extension and hyperextension. REE, right elbow extension.

- Reporting of reasons for missingness (eg, post-operative restrictions) was variable by joint, but reasons were not commonly recorded for joint/side measurements with high missingness (eg, left elbow extension; **Figure 2**)

Figure 2. Missing measures over time and whether a reason for missingness was provided for A) left elbow flexion and B) left elbow extension



Data are for the full cohort (n = 1989). Reasons for missingness included "acute bleed," "post-operative restrictions," and "other medical reason."

- Only approximately 50% (n = 1072) of the overall population had ≥1 complete ROM assessment recorded in their history (**Table 3**)
  - A majority of those with complete ROM assessments had only 1 or 2 complete ROM assessments, with 38% (n = 408) having only 1 complete assessment

Table 3. Number of individuals in the full cohort (n = 1989) with complete joint ROM assessments

Number of complete assessments	Number of individuals	Cumulative count
15	1	1
13	1	2
12	1	3
11	3	6
10	8	14
9	11	25
8	24	49
7	32	81
6	42	123
5	61	184
4	79	263
3	149	412
2	252	664
1	408	1072

Complete ROM assessments included recorded flexion/plantarflexion and extension/dorsiflexion measurements on both sides for the ankle, knee, hip, and elbow, as well as shoulder flexion on both sides. ROM, range of motion.

- Overall, the observed missing data on joint ROM in the ATHNdataset prevented further analyses of joint mobility using these data

Conclusions

- Systematic missingness appears to exist in ROM data recorded in the ATHNdataset. The impact of missingness on ROM, combined with potential intra-patient and intra-therapist variability, limits the ability to describe generalizable longitudinal changes in ROM
  - ROM data collection is labor intensive;[2] usability of tools is equally important as comprehensive data collection
- Specific joints without high missingness may still provide opportunities for studying ROM over time, only without the ability to assess "overall joint health" as a function of ROM in elbows, knees, and ankles
- This assessment highlights the importance of full data recording in the ATHNdataset to facilitate future research, as well as potential limitations when analyzing historically collected data
- The ATHN Transcends protocol aims to improve data completeness and quality in general, with the goal of addressing some of the current limitations of the ATHNdataset

References

1. American Thrombosis and Hemostasis Network. https://www.athn.org/what-we-do/national-projects/athndataset.html Accessed Sept 9, 2022. 2. Soucie, JM, et al. *Haemophilia*. 2011;17(3):500–7.

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Disclosures

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