H:\EGS\Article-EGS\JCEMrevised4\JCEMrevisedFigures\vdStraaten_jc.2019-01437_SuplFig1A.tif

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**Supplemental figure 1.** Correlation and agreement between EGS and EMS. A: Correlation between EGS and EMS obtained in babies with typical and atypical genitalia. B: Bland-Altman plot to assess agreement between EGS and EMS. The difference between EGS and EMS against the mean of EGS and EMS.

|  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- |
| **EMS** | **outcomes** | **sub scores** | **EGS** | **outcomes** | **sub scores** |
| **Scrotal fusion** | **Y/N** | **3 / 0** | **L/S fusion** | **Fused / Posterior fusion / Unfused** | **3 / 1.5 / 0** |
| **Micropenis** | **N/Y** | **3 / 0** | **Genital**  **tubercle**  **length** | **>31 / 26-30 /21-25 / 10-20 / <10** | **3 / 2.5 / 1.5 / 1/ 0** |
| **Urethral meatus** | **Normal/Distal**  **Mid / Proximal** | **3 / 2**  **1 / 0** | **Urethral**  **meatus** | **Top GT / Coronal, Glandular**  **Along GT / GT base / LS/ Perineal** | **3 / 2.5**  **2 / 1.5 / 1 / 0** |
| **Right gonad** | **LS /inguinal /Absent** | **1.5 / 1 / 0** | **Right gonad** | **LS/ Ing-LS / Inguinal / Impalpable** | **1.5 / 1 / 0.5 / 0** |
| **Left gonad** | **LS/inguinal / Absent** | **1.5 / 1 / 0** | **Left gonad** | **LS/ Ing-LS / Inguinal / Impalpable** | **1.5 / 1 / 0.5 / 0** |
| **total score** | **12** | | **total score** | **12** | |

**Supplemental table 1.** Modification of the “External Masculinization Score” into the “External Genitalia Score” in order to describe the full spectrum of naturally occurring phenotypic variation in external genitalia. The EGS and EMS describe the same phenotypic features at 5 anatomical landmarks of the genitalia: degree of labioscrotal fusion, length of the genital tubercle, position of the urethral meatus, and location of the right and left gonad. The categorical scales of EGS are more refined as compared to EMS, genital tubercle length has become a gradual scale. The final score is the sum of points allocated to feature 1-5, with a maximum of 12 for both scores.

Abbreviations: EMS: External Masculinization Score, EGS: External Genitalia Score. GT: genital tubercle, LS : Labio/scrotal, Ing-LS : Inguino-Labio/scrotal

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Inter-observer Intraclass Correlation** | | | | | | | |
| **Typical**  **Genitalia** | C N | ICC | 95% CI | **Atypical genitalia** | C N | ICC | 95% CI |
| EGS | C 8 35 | 1 | 0.99-1.0 | EGS | C 1 10  C 7 17  C 8 39 | 0.95  0.93  0.80 | 0.83-0.99  0.83-0.98  0.65-0.89 |
| Prader | C 8 35 | 1 | 1.0-1.0 | Prader | C 1 10  C 7 17 | 0.92  0.58 | 0.71-0.98  0.16-0.82 |
| EMS | C 8 35 | 1 | 1.0-1.0 | EMS | C 1 10  C 7 17  C 8 39 | 0.88  0.83  0.71 | 0.62-0.97  0.59-0.93  0.52-0.84 |
| **FEMALE**  AGDl | C 8 12 | 0.85 | 0.55-0.95 |  |  |  |  |
| AGDu | C 8 12 | 0.76 | 0.37-0.92 |  |  |  |  |
| **MALE**  AGDl | C 8 23 | 0.86 | 0.70-0.94 | AGDl | C 1 6  C 7 17  C 8 39 | 0.92  0.87  0.87 | 0.50-0.99  0.68-0.95  0.77-0.93 |
| AGDu  Genital tubercle length | C 8 23  C 8 23 | 0.52  0.77 | 0.13-0.76  0.53-0.90 | AGDu | C 1 5  C 7 17  C 8 39 | 0.97  0.87  0.93 | 0.81-1.0  0.68-0.95  0.86-0.96 |
|  |  |  |  | Genital tubercle length | C 1 8  C 7 17  C 8 39 | 0.89  0.57  0.90 | 0.59-0.98  0.17-0.82  0.82-0.97 |
| **Intra-observer Intraclass Correlation** | | | | | | | |
| **Typical**  **Genitalia** | C N | ICC | 95% CI | **Atypical**  **genitalia** | C N | ICC | 95% CI |
| **FEMALE**  AGDl | C 8 12 | 0.89 | 0.67-0.97 |  |  |  |  |
| AGDu | C 8 12 | 0.85 | 0.57-0.96 |  |  |  |  |
| **MALE**  AGDl | C 8 23 | 0.93 | 0.84-0.97 | AGDl | C 1 6  C 8 39 | 0.99  0.84 | 0.92-1.0  0.73-0.91 |
| AGDu  Genital tubercle length | C 8 23  C 8 23 | 0.87  0.82 | 0.71-0.94  0.62-0.92 | AGDu | C 1 5  C 8 39 | 0.96  0.97 | 0.75-1.0  0.95-0.98 |
|  |  |  |  | Genital tubercle length | C 1 6  C 8 39 | 0.99  0.97 | 0.99-1.0  0.97-1.0 |

**Supplemental table 2.** Inter-observer ICC for EGS, EMS, PS, AGDl and AGDu, genital tubercle length and intra-observer ICC for EGS, EMS, PS in babies with typical and atypical genitalia Inter- and intra-observer ICC were calculated using Single Rating, Absolute agreement, 2-way Random-effect Model. Abbreviations: EGS: External Genitalia Score, EMS: External Masculinization Score, ICC: Intraclass correlation coefficient, CI: confidence interval, C1- 8: center 1- 8, AGD: anogenital distance, AGDu (AGDupper): measured from the centre of the anus to the anterior base of the genital tubercle, and AGDl (AGDlower) is measured from the centre from the anus to the base of the labio/scrotal border.

|  |  |  |
| --- | --- | --- |
| Characteristics for babies with typical genital phenotype |  | N |
| Total | Participant | 867 |
| Sex | female | 431 |
|  | male | 436 |
| Gestational age (weeks) | <28 | 44 |
|  | 28-32.9 | 62 |
|  | 33-36.9 | 75 |
|  | >37 | 686 |
| Birth weight (gram) | <1000 | 48 |
|  | 1000-1500 | 36 |
|  | 1500-2500 | 88 |
|  | 2500-4000 | 619 |
|  | >4000 | 76 |
| SGA or AGA | AGA | 811 |
|  | SGA | 56 |
| Age (months) | 0-1 | 507 |
|  | 1-5 | 235 |
|  | 6-11 | 87 |
|  | 12-24 | 38 |
| Study center | Center 1 | 222 |
|  | Center 2 | 291 |
|  | Center 3 | 104 |
|  | Center 4 | 122 |
|  | Center 5 | 90 |
|  | Center 6 | 38 |
| Characteristics for infants with atypical genital phenotype |  | N |
| Total | participant | 111 |
| DSD | 46,XY DSD | 46 |
|  | Sex Chromosome DSD | 10 |
|  | Mild, non-specific undermasculinization\* | 45 |
|  | 46,XX DSD | 10 |
| Study center | Center 1 | 36 |
|  | Center 6 | 11 |
|  | Center 7 | 17 |
|  | Center 8 | 47 |

**Supplemental table 3.** Participant characteristics

Multicenter study for validation of EGS and correlation between EGS and AGD-ratio

Abbreviations: SGA: small for gestational age, AGA: appropriate for gestational age

\* Mild, non-specific undermasculinization relates to male babies with isolated cryptorchidism or mild hypospadias

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  | | | EGS | EMS |
| Spearman's rho | Ratio AGDl/u | Correlation Coefficient | 0.19 | 0.88 |
| Sig. | <0.05 | 0.09 |
| N | 243 | 243 |
|  | AGDl | Correlation Coefficient | 0.29 | 0.2 |
|  |  | Sig. | <0.05 | <0.05 |
|  |  | N | 243 | 243 |
|  | AGDu | Correlation Coefficient | 0.23 | 0.23 |
|  |  | Sig. | <0.05 | <0.05 |
|  |  | N | 243 | 243 |

**Supplemental table 4.** Correlation between ratio AGDl/u, AGDl, AGDu and EGS and EMS in neonates with typical male phenotype

Abbreviations: AGD: anogenital distance, AGDu (AGDupper): measured from the centre of the anus to the anterior base of the genital tubercle, and AGDl (AGDlower): measured from the anus to the base of the labio/scrotal border. AGDl/u: lower /upper AGD ratio