Multidimensional outcome assessment in cerebral palsy children: “the STIC AQM project”

Viehweger E. MD PhD
Haumont T. MD PhD, Presedo A. MD, De Lattre C. MD, Loundou A. PhD,
Simeoni MC. MD PhD,
and the VARAX Study Group

Pediatric Orthopaedic Department
Pr. G. BOLLINI
Timone Children’s Hospital
Research Unity EA 3279 Health Evaluation and Perceived Health
Public Health Department, Faculty of Medicine
Marseille, France
Cerebral Palsy

- Static cerebral pathology
- Cerebral damage with non-progressive pathology of posture and movement
- Variable etiologies
- Associated to epilepsy, speaking problems, vision and hearing and cognitive involvement
Quality for all health care participants

- legal framework
- Benefit for the patient
  - Comparison before / after treatment
- Objectives
- Proof of treatment efficiency
  - reliable
  - limits of personal experience
  - level of proof
- Evidence-based Medicine
- Professional recommendations
ICF: International Classification of Functioning, Disability and Health (WHO, 2001)

- Multidimensional evaluation

- requirements of ICF (2001)
ICF: International Classification of Functioning, Disability and Health (WHO, 2001)

ICF Conceptual Interaction

Health Problem (trouble/disease)

Deficiency (fonction/structure)  Activities (activity limitation)  Participation (restriction of participation)

Environmental factors  Personal factors
ICF in practice

- Evaluation of the needs
- Evaluation of the results
- Comparison of different treatments
- Implication of consumer
- Habitudes of use
- Performances in services
  - Results
  - Cost – efficiency
- Electronic files
- Clinical terminology
for all health practitioners (physiatrists, surgeons, prothesists...)

- global outcome assessment: international standard
- Multi-factorial
  - Clinical variables
  - Tools
  - Scores
- Multidisciplinary: practice

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</thead>
</table>
Spasticity
Selectivity
Coordination
Spasticity
Selectivity
Coordination
Abnormal Forces
Growth
Bony deformities
Contractures
1° Lesion CNS
2° Lesions
3° Lesions

Introduction
Gait Analysis
+++
EVALUATION

Hägglund G  JPO July 2005

209 children

40%

Bony/muscle procedures

<table>
<thead>
<tr>
<th>Year of birth</th>
<th>Proportion (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1990–1991</td>
<td>45</td>
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<tr>
<td>1992–1993</td>
<td>35</td>
</tr>
<tr>
<td>1994–1995</td>
<td>15</td>
</tr>
</tbody>
</table>

Introduction
Concept Study Discussion Conclusion
EVALUATION

Adaptation therapy

History of CP treatment

Now

J GAGE

F MILLER

MD SUSSMAN

No toxin treatment

« late » toxin treatment

MLS

0 5

Multiple bony deformities
Multiple "soft tissue contractures"

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EVALUATION

Identification of anomalies

Tools

| Introduction | Concept | Study | Discussion | Conclusion |
|--------------|---------|-------|------------|------------|------------|
|              |         |       |            |            |            |
Understanding Déformations osseuses

Introduction

Concept Study Discussion Conclusion
EVALUATION

diplegic
diplegic

BEFORE

AFTER

No clinical "normalisation"

Better function ?

Better quality of life ?

Introduction | Concept | Study | Discussion | Conclusion
• no homogenous criteria for orthopaedic decision making in CP children
• decision making process more complex
• motor impairment, tonus modification, severity of involvement
• analysis of complex quantitative 3-D gait data
Multidimensional

Goldberg’s Model *(JPO 1994)*

- Organic function and anatomical structure
- Technical
- Gait Analysis
- Functional Scores
- HRQL
- Cost
- Environmental factors
- Evaluation strategies, therapeutics
  - conceptual analysis
  - comparison strategies

Introduction | Concept | Study | Discussion | Conclusion
---|---|---|---|---

A French multicenter study

Usefulness of Gait Analysis in Evaluation of Disabled Patients with Walking Potential

Grant DHOS/OPRC/2003/167; April 4th 2003
“Programme de soutien des innovations diagnostiques et thérapeutiques coûteuses 2003”
French Ministry of Health, Family and Disabled Persons.
Concepts to know?

- Age
- Gender
- Pain
- CP Type
- Handicap
  - IQ
  - visual or hearing deficit
  - ...
- Gait Pattern
Sagittal Gait Pattern

Sagittal gait patterns in spastic diplegia

VOL. 86-B, No. 2, MARCH 2004

Introduction

Concepts

Study

Discussion

Conclusion
CP Classifications

- **Monoplegia**
- **Paraplegia**
- **Hemiplegia**
- **Triplegia**
- **Quadriplegia**
- **Diplegia**

**Topographical**

**Physiological**

- Athetoid (permanent movements, uncontroled)
- Spastic +++
- Rigidity
- Ataxic (poor equilibrium, falls, ...)
- Tremor

**Etiological**

- **Prenatal (70%)**
  - Infection, anoxia, toxic, vascular, Rh pathology, genetic, cerebral malformation
- **At birth (5-10%)**
  - Anoxia, traumatism, metabolic
- **After birth**
  - Traumatism, infection, toxic

**Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers**

*Developmental Medicine & Child Neurology 2000, 42: 816-824*
EVALUATION

Gross Motor Function Classification System

Classifying Cerebral Palsy
[On the Other Hand]

Graham, H. Kerr MD, FRCS(Ed), FRACS

Palisano Dev Med Child Neuro 1997

Introduction Concepts Study Discussion Conclusion
Functional

Gross Motor Function Measure (GMFM)
(EMFG validated in French, Escale team (Lyon))

- 5 dimensions
  - A: Lying and rolling
  - B: Sitting
  - C: Crawling and kneeling
  - D: Upright
  - E: Walking, running and jumping
- total score and Score D+E
- with / without orthosis

Introduction | Concepts | Study | Discussion | Conclusion
Functional Assessment Questionnaire (FAQ)

10-level FAQ

Mean cardiac frequency at 5th walking minute (beats/minute)

\[ \text{EEI} = \frac{\text{Walking velocity (m/min.)}}{} \]

| Introduction | Concepts | Study | Discussion | Conclusion |
Technical

- Know what you want to measure
- Clinical / Research question

- Extraction specific data
  - Kinematic/gait cycle events
  - Modification force/moment
  - spatio-temporal parameters

- Use of Indexes extracted out of Gait Analysis
  - Gillette Gait Index (GGI)
“Normalcy Index (NI)”
= Gillette Gait Index (GGI)

M. Schwartz, PhD

- quantitative measure of global gait pathology
- 16 kinematic parameters (experience, conveniance)
- multivariated principal component analysis
- Correlations with severity (Type, MRI, …)
- Validation, Test / evaluation tools
- Normal 15

Introduction in France
(E. Viehweger MD, PhD)
Effect of Diagnosis

N=145  Hemiplegia
N=331  Diplegia
N=82   Quadriplegia

Knee flexion at initial contact

Introduction  Concepts  Study  Discussion  Conclusion
Concepts to know

- **Quality of life**
  - Generic (different pathologies) / Specific

- **Construction / administration modus**
  (patient, experts, family...)

- **Validated tools**
  (validity, reliability, sensibility to change, acceptability)

Need of transcultural validation ++++
Interest of measuring QL in CP patients

- Complementary to clinical measures, symptoms
- Effect of the pathology and its treatments
  - Physical
  - Psychological
  - Social
- Perception of the patient
### 18 Instruments in CP children

#### Generic QL used in CP children
- VSP-A
- KIDSCREEN
- CHQ
- CHIP
- TACQOL
- Exeter HRQL
- PedsQL
- GCQ
- AUQUEI
- DHP-A
- KidlQol
- KINDL

#### Specific CP or neuromuscular QL
- DISABKIDS
  - « Cerebral palsy » module
  - CP-QOL
  - FMH (activity and participation)
  - Life-H (activity and participation)
  - LSIA (Life satisfaction Index of Adolescents)
  - HUI-3

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<th>Conclusion</th>
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</thead>
<tbody>
<tr>
<td>Instrument</td>
<td>Versions</td>
<td>Principal Reference</td>
<td>Original language</td>
<td>Dominant point of view</td>
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<tr>
<td>------------</td>
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<td>------------------------</td>
</tr>
<tr>
<td>AUQUEI Child 4-12</td>
<td>Manificat S. (1997)</td>
<td>France</td>
<td>Children</td>
<td>27-33</td>
</tr>
<tr>
<td>PEDSQL Pediatric QL Inventory</td>
<td>Child 5-7 Child 8-12 Child 13-18 Parent Version</td>
<td>Varni JM. (1999)</td>
<td>USA</td>
<td>Children Parents Health professional Literature</td>
</tr>
<tr>
<td>VSP-A Vécu et Santé Perçue de l’Adolescent</td>
<td>VSPA (11-18) VSPAe (8-10) VSPA12 Parent Version</td>
<td>Simeoni MC. (2000)</td>
<td>France</td>
<td>Children Adolescents</td>
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<tr>
<td>DISABKIDS Child 8-16 Child 4-7 Parent Version</td>
<td>Bullinger M. (2002)</td>
<td>European Project</td>
<td>Children Parents Health professional Literature</td>
<td>37/ 12 + CP : 7</td>
</tr>
<tr>
<td>DHP-A Child 13-18</td>
<td>Vo TX. (2005)</td>
<td>UK</td>
<td>Experts Physicians</td>
<td>17</td>
</tr>
</tbody>
</table>
« European » Questionnaires
(multilingual, multicultural)

KIDSCREEN

DISABKIDS Chronic Generic Module

DISABKIDS Disease specific Module

Epilepsy  Diabetes  Rhum. arthritis  CP  ...

Introduction  Concepts  Study  Discussion  Conclusion
DISABKIDS development

- Literature review
- Focus groups and interviews
- Item selection
- Translations
- Pilot study (CP : 21)
- Field study
- Implementation study
Development of a condition-specific measure of quality of life for children with CP: empirical thematic data reported by parents and children (CPQoL)


- Interviews to identify principal themes which define QoL in CP children
- CP children and parents
- 42 families (Melbourne register, Australia) (≠ age, functional status, socio-economical status, rural/urban)
- 28 participating families
  - Parents: 16 of children 4-8 y + 12 of children 9-12 y
  - Children:
    - 5 of 12 children 9-12 y (GMFCS Palisano I-II)
    - 5 not able to participate (GMFCS Palisano III-IV-V)
    - 2 parental chosen timing not compatible to child’s presence
VSP-A = Vécu et Santé perçu de l’Adolescent

- Generic
- Adolescent (11-18 y), Children (6-10 y), Parent
- Validation +++
- 38 – 39 – 46 Questions
- QL Index et 10 profiles
  - energy – vitality
  - psychological well being
  - physical well being
  - relation with parents, friends, caregivers, teachers
  - scolar work
  - leasure
  - Self esteem

Scores of domains from 0 to 100 (0=worst QL, 100=excellent QL)

Marie-Claude Simeoni, Stéphane Robitail, Pascal Auquier
EA 3279 : Evaluation hospitalière et santé perçue
Faculty of Medicine, Marseille

| Introduction | Concepts | Study | Discussion | Conclusion |
A French multicenter study

Usefulness of Gait Analysis in Evaluation of Disabled Patients with Walking Potential

Grant DHOS/OPRC/2003/167
April 4th 2003
“Programme de soutien des innovations diagnostiques et thérapeutiques coûteuses 2003” French Ministry of Health, Family and Disabled Persons.
• 3-year prospective multicenter study
• 6 to 18 years
• Demographic data
• CP Type
• Pain
• Orthosis, assistive device
• Homogenous clinical exam (Delphi procedure)

Introduction
Concepts
Study
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Conclusion

- Handicap
  - cognitive
  - visual or hearing deficit
  - Gross Motor Function (GMFCS): 1 - 4

- Gait Pattern

Testing range of motion in cerebral palsy
E. Viehweger, C. Béard, A. Berruyer, M.-C. Simeoni, Groupe Varax
Annales de réadaptation et de médecine physique 50 (2007) 258–265
Outcome evaluation tools

- Technical: GGI (gait analysis)
- Functional: GMFM D+E, 10-level FAQ, EEI

\[
EEI = \frac{\text{Mean cardiac frequency at 5th walking minute (beats/minute)}}{\text{Walking velocity (m/min.)}}
\]

- HRQL: VSP-A
  (children, adolescents, parents)
All gait labs
with own database of “normals”

<table>
<thead>
<tr>
<th>Center</th>
<th>Le Mans</th>
<th>Lyon</th>
<th>Marseille</th>
<th>Nancy</th>
<th>Paris</th>
<th>Toulouse</th>
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<tbody>
<tr>
<td><strong>Number</strong></td>
<td>29</td>
<td>30</td>
<td>27</td>
<td>30</td>
<td>32</td>
<td>36</td>
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<tr>
<td><strong>Mean GGI</strong></td>
<td>15,7</td>
<td>15,4</td>
<td>15,4</td>
<td>15,7</td>
<td>15</td>
<td>15,1</td>
</tr>
</tbody>
</table>
Common database
160 diplegic cerebral palsy children

- 11.0 years (range 4.3 to 18.5 years; SD 3.2 years)
- 95 boys (59.4%) and 65 girls (40.6%)
- 11.0 % dependant ambulators (crutches, K-walkers)
- 25.0 % orthosis
- 11.6% slight, 3.4% moderate cognitive deficiency
GMFCS

GMFCs 1: 19
GMFCs 2: 56
GMFCs 3: 63
GMFCs 4: 22

Rodda

Asymmetric: 18
Crouch: 51
Apparent equinus: 16
Jump Gait: 35
True equinus: 40

Introduction  Concepts  Study  Discussion  Conclusion
160 spastic diplegic CP

GGI - GMFCS

GMFCS 1: 184,29±190,87
GMFCS 2: 210,60±129,68
GMFCS 3: 291,61±175,56
GMFCS 4: 494,71±241,13

Introduction
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160 spastic diplegic CP

GGI - Rodda

<table>
<thead>
<tr>
<th>Rodda 1</th>
<th>Rodda 2</th>
<th>Rodda 3</th>
<th>Rodda 4</th>
<th>Rodda 5</th>
</tr>
</thead>
<tbody>
<tr>
<td>246.7 +/- 146.5</td>
<td>274.4 +/- 229.7</td>
<td>314.7 +/- 309.4</td>
<td>276.6 +/- 174.6</td>
<td>246.8 +/- 115.5</td>
</tr>
</tbody>
</table>

Introduction | Concepts | Study | Discussion | Conclusion
Correlations between quantitative/qualitative outcome tools

GMFM

GGI

FAQ

EEI

(p<0.0001, Kendall’s Tau)

VSP-A

VSP-Ae

Introduction | Concepts | Study | Discussion | Conclusion

--- | --- | --- | --- | ---
GMFCS

- significant differences: GGI, GMFM D+E, FAQ, EEI

  - increasing gait pathology (GGI)
    = more involved gross motor function

  - more involved gross motor function
    = decreasing function (GMFM, FAQ)
    = increased EEI

Rodda

- no significant differences
Quality of Life (VSP-A)

109 patients (68.1%) : 43 adolescents, 66 children
80 VSP-A parent questionnaires (50%)
Introduction

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Conclusion
VSP-A adolescents 11 - 18 y (general population versus CP)

QoL Index

General population
CP adolescents

Introduction
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Study
Discussion
Conclusion
3 treatment groups

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<thead>
<tr>
<th>Treatment</th>
<th>N</th>
<th>Percentage</th>
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<tbody>
<tr>
<td>Physiotherapy</td>
<td>37</td>
<td>23%</td>
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<tr>
<td>Botulinum toxine</td>
<td>41</td>
<td>26%</td>
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<tr>
<td>Surgery</td>
<td>82</td>
<td>51%</td>
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<table>
<thead>
<tr>
<th>Age *</th>
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<tbody>
<tr>
<td>Physiotherapy</td>
<td>10.83 ± 3.11</td>
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<tr>
<td>Botulinum toxine</td>
<td>8.79 ± 2.69</td>
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<tr>
<td>Surgery</td>
<td>11.05 ± 3.21</td>
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<thead>
<tr>
<th>Assistive device</th>
<th></th>
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<tbody>
<tr>
<td>Physiotherapy</td>
<td>41.5%</td>
<td></td>
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<tr>
<td>Botulinum toxine</td>
<td>47.8%</td>
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<tr>
<td>Surgery</td>
<td>57.3%</td>
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<th>Orthosis*</th>
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<td>Physiotherapy</td>
<td>41.5%</td>
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<tr>
<td>Botulinum toxine</td>
<td>29.8%</td>
<td></td>
</tr>
<tr>
<td>Surgery</td>
<td>13.5%</td>
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</tr>
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- Concepts
- Study
- Discussion
- Conclusion
Evaluation at inclusion – 9 months

GGI

GMFM D+E

EEI

Physiotherapy: NS

Botulinum toxine: S

Surgery: S
Variations: clinically significant?

GMFM D+E

Introduction
Concepts
Study
Discussion
Conclusion
Variations: clinically significant?

10-level FAQ
Variations: clinically significant?

EEI

![Graph showing variations in EEI with specific values marked for PT, Btx-A, Surgery, and All categories.](image)

- PT: +0.16
- Btx-A: 0.54
- Surgery: +0.98
- All: +0.31
Variations: clinically significant?

GGI

- 25.4%
- 29.4%
- 21.2%
+ 4.6%

Introduction  Concepts  Study  Discussion  Conclusion
– Use of Classifications – Patient groups
  • Commun language

– « homogenous » evaluation
  • multidisciplinary
  • comprehension physiopathology
  • therapeutic objectives

– Diffusion functional evaluation tools
  • GMFM, EEI, FAQ, ...

– Integration technical evaluation
  • discriminatory abilities

– Quality of Life :
  • multicenter studies
  • new perspectives
Quality of Life (VSP-A)

- no discrimination GMFCS level
- no correlation QoL scores and GGI, GMFM, FAQ, EEI
- questionnaire (dominant domains, validation)
- differences self / proxy evaluation
- disability paradox
Quality of Life (VSP-A)

Content Comparison

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<tr>
<th>Field</th>
<th>Domain</th>
<th>Examples</th>
<th>PedsQL</th>
<th>Kidscreen</th>
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<tr>
<td>Physical</td>
<td>Physical activity</td>
<td>Sports, physical exercise</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Restriction of activities</td>
<td>Limitation walking, running, sitting, climbing stairs</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Physical symptoms</td>
<td>pain, headache</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Sensation</td>
<td>Full fitness, to feel resistant</td>
<td></td>
<td></td>
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<tr>
<td>Psychological</td>
<td>Negative thinking</td>
<td>To feel worried, sad</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>Positive emotions</td>
<td>To feel nice, happy</td>
<td>+</td>
<td></td>
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<tr>
<td></td>
<td>Self esteem</td>
<td>To be satisfied with its appearance, of its accomplishments</td>
<td></td>
<td>+</td>
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<tr>
<td></td>
<td>Cognitive functioning</td>
<td>Difficulties to concentrate, to learn</td>
<td></td>
<td>+</td>
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<tr>
<td></td>
<td>Behavior</td>
<td>Compared to age, contest</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social</td>
<td>Friends</td>
<td>Relations, influence</td>
<td>+ (neg)</td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>School</td>
<td>Relations teachers, work</td>
<td></td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>Family</td>
<td>Activities, comprehension, attention, help, protection</td>
<td></td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>Isolation, social integration</td>
<td>To feel lonely, excluded</td>
<td></td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>Financial resources</td>
<td>Sufficient money to follow up its friends</td>
<td></td>
<td>+</td>
</tr>
</tbody>
</table>

Adapted according to Rajmil L. et al. Journal of Adolescent Health 2004

| Introduction | Concepts | Study | Discussion | Conclusion |
|--------------|----------|-------|------------|------------|------------|
Quality of Life (VSP-A)

- no discrimination GMFCS level
- no correlation QoL scores and GGI, GMFM, FAQ, EEI
- questionnaire (dominant domains, validation)
- differences self / proxy evaluation
- disability paradox


- awareness of pathology and future implication?
Conclusions

- good concordance technical / functional measures
- GGI best to describe changes in time
- lack of correlation with QoL
- choice of questionnaire

Generic questionnaire *(ex: KIDSCREEn)*

+ specific *(DISABKIDS)*

+ Participation *(Life-H)*
Conclusions

Objective according to severity

**GAS**

- Autonomy outside
- Esthetic
- Interior autonomy
- Hand function
- Speech
- Survival
- Nursing
- No pain

**Professional recommendations**

- Tone treatment (toxine)
- Maintain muscle force
- Weight control
- Orthopaedic Surgery (lever arms)

- Tone treatment (toxine, pump)
- Maintain muscle force
- Weight control
- Orthopaedic Surgery (lever arms)

- Nutrition (gastrostomy)
- Tone treatment (toxine, pump)
- Maintain muscle force
- Orthopaedic Surgery

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Thank you