Pain in the child with severe neurological impairment (SNI)

Catherine Diskin

SickKids®

Complex Care

UNIVERSITY of TORONTO

I (we) would like to begin by acknowledging the land on which SickKids operates. For thousands of years it has been the traditional land of the Huron-Wendat and Petun First Nations, the Seneca, and most recently, the Mississaugas of the Credit River. Today, Toronto is home to Indigenous Peoples from across Turtle Island. SickKids is committed to working toward new relationships that include First Nations, Inuit, and Métis peoples, and is grateful for the opportunity to share this land in caring for children and their families.

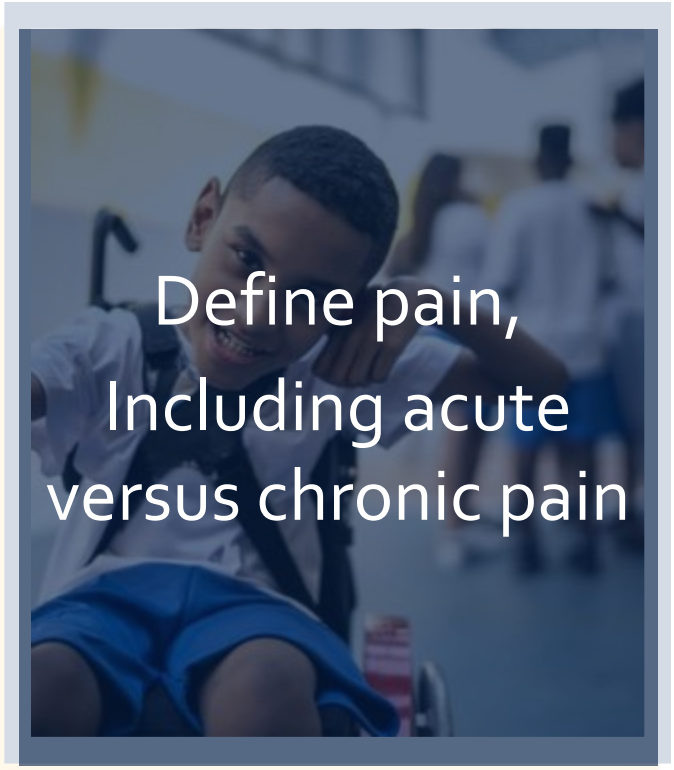
Art by Emily Kewageshig

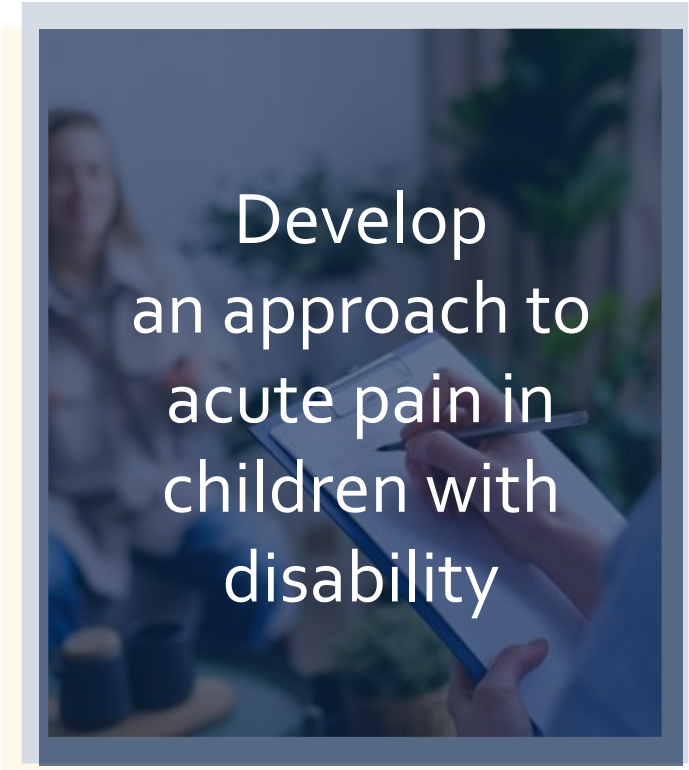
SickKids®

Mitigating potential bias

We have no commercial interests to disclose.

Learning Objectives





Children with medical complexity and neurological impairment

Child with medical complexity "substantial family-identified service needs, chronic conditions, functional limitations and high health care use".[1]

Neurologic impairment (NI): " neurologic condition primarily arising from the central and/or peripheral nervous systems.. at least 12 months AND results in systemic and/or multisystem physiologic impairment ..."[2]

1. Cohen E, Kuo DZ, Agrawal R, et al. Children With Medical Complexity: An Emerging Population for Clinical and Research Initiatives. *Pediatrics*. 2011
2. Berry JG, Poduri A, Bonkowsky JL, et al. Trends in resource utilization by children with neurological impairment in the United States inpatient health care system: a repeat cross-sectional study. *PLoS Med*. 2012

PAIN

Pain is “an unpleasant sensory and emotional experience associated with, or resembling that associated with actual or potential tissue damage”

Additional six key notes addended to definition

(International Association for the Study of Pain)

6 key notes about Pain

- Pain is always a **personal** experience
- Pain and nociception are different phenomena
- Through their **life experiences**, individuals learn the concept of pain
- A person's report of an experience as pain should be **respected**
- Although pain usually serves an adaptive role, it may have **adverse effects** on function and social and psychological well-being
- Verbal description is only one of several behaviours to **express pain**

International Association for the Study of Pain, 2020

Acute versus chronic pain

ACUTE

- Caused by tissue inflammation or injury (nociceptive)
- Hours – days
- Diagnostic tests can be helpful
- Resolution expected after treatment and healing

CHRONIC

- Variety of categories – musculoskeletal, neuropathic, visceral
- Persistent; > 3months by definition
- Diagnostic tests often unhelpful
- Focus is often on pain control

Hauer J, Houtrow AJ; SECTION ON HOSPICE AND PALLIATIVE MEDICINE, COUNCIL ON CHILDREN WITH DISABILITIES. Pain Assessment and Treatment in Children With Significant Impairment of the Central Nervous System. Pediatrics. 2017 Jun;139(6):e20171002. doi: 10.1542/peds.2017-1002. PMID: 28562301.

Agitation, Irritability, Neuroirritability

- **What do we mean?** Descriptive terms: Unpleasant state of arousal, the absence of calm

- **What can cause agitation?** Unmet need, emotional state, acute illness, drug, pain

- More than one cause is possible

US Department of Health and Human Services; National Institutes of Health; National Cancer Institute. Common Terminology Criteria for Adverse Events.

Pain in children with SNI is a common problem

- Up to 42% of children with SNI **daily** (2)
- Parents of children with a G-tube requiring the greatest number of medications report the highest pain frequency (3)
  - Pain is typically described as moderate to severe (4)
- When a child experiences pain, it impacts their quality of life including sleep (5)

Challenges in the assessment of pain in children with SNI

- False assumptions
- Communication and understanding of pain
- Limited understanding of pain in children with SNI (8)
- Difficulty identifying source of pain

Hauer JM. Pain in Children with Severe Neurologic Impairment: Undoing Assumptions. *JAMA Pediatr*. 2018;172(10):899-900. doi:10.1001/jamapediatrics.2018.1531

Pain is a natural part of the patient's underlying condition

Pain rarely needs treatment

A non-verbal child cannot communicate they are in pain

Challenges: False assumptions

Motor, cognitive, and communication difficulties make accurate assessment of pain difficult

Can exhibit a wide range of pain behaviours

Hadden KL, et al, 2002; Chen-Lim et al, 2012;Oberlander et al, 1999; Hunter et al, 2016, Hunt et al 2003

Challenges: Communications

LIMITED RESEARCH FOCUSED ON THIS CLINICAL PHENOMENON

ABSENCE OF CLINICAL GUIDELINES

CONTRIBUTES TO HIGH VARIABILITY BETWEEN PHYSICIANS IN THE TREATMENT OF PAIN/IRRITABILITY OF UNKNOWN ORIGIN IN CHILDREN WITH SNI (8)

Challenges: Limited knowledge

May not identify the source of pain

Sources of pain can also be pain behaviours

Dystonia

Feeding intolerance

Challenges: Identification of source of pain

Developing an approach to a child with SNI experiencing pain

An approach to pain in child with SNI

1. Careful history
2. Thorough physical examination
3. Prudent clinical testing, directed by history and examination
4. Early consideration and instigation of pharmacological and non-pharmacological treatment of pain

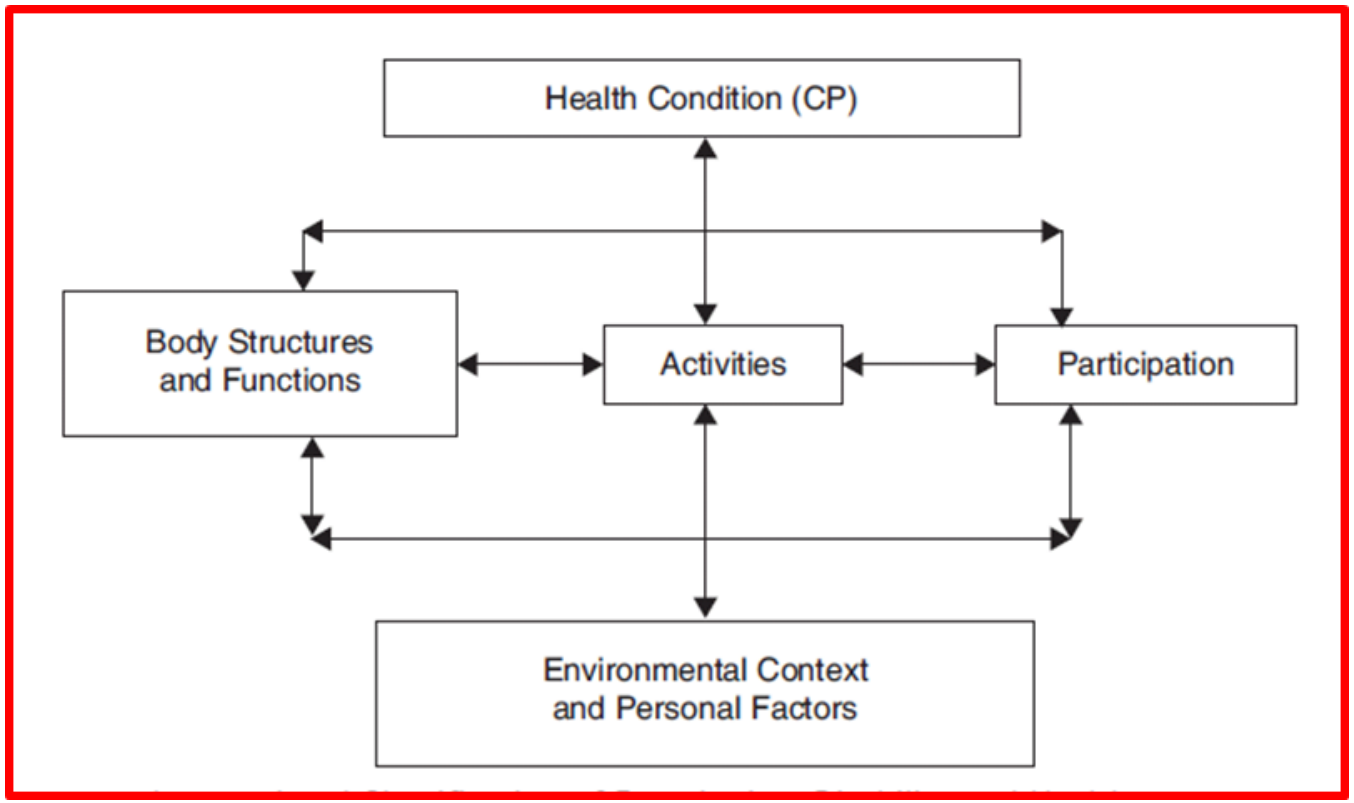
An approach to pain in child with SNI: Careful history

History

- Consider using the International Classification of Functioning, Disability and Health Model to direct a comprehensive (pain) assessment.

- Causes of pain in the general population also occur in children with SNI

- We have the potential to cause more pain



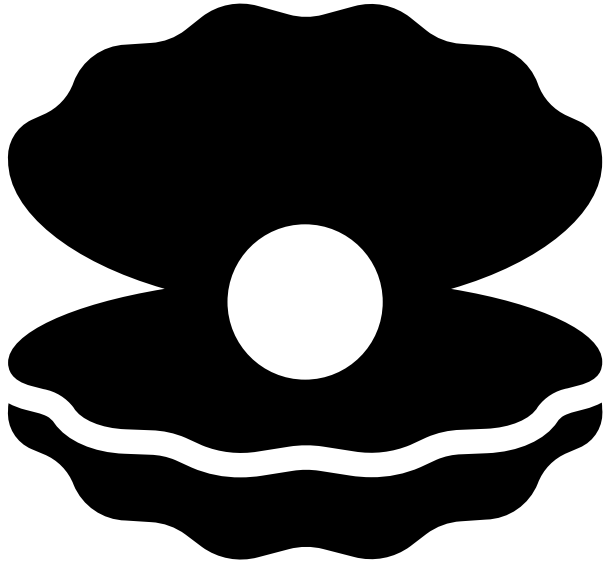
World Health Organization 2001. The International Classification of Functioning, Disability and Health (ICF). Geneva: WHO. http://www.who.int/classifications/icf/en/

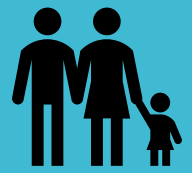
Clinical pearl

A child with SNI can communicate in many ways.

Verbal self-reporting including use of yes/no responses, augmentative communication strategies or non-verbal communication strategies.

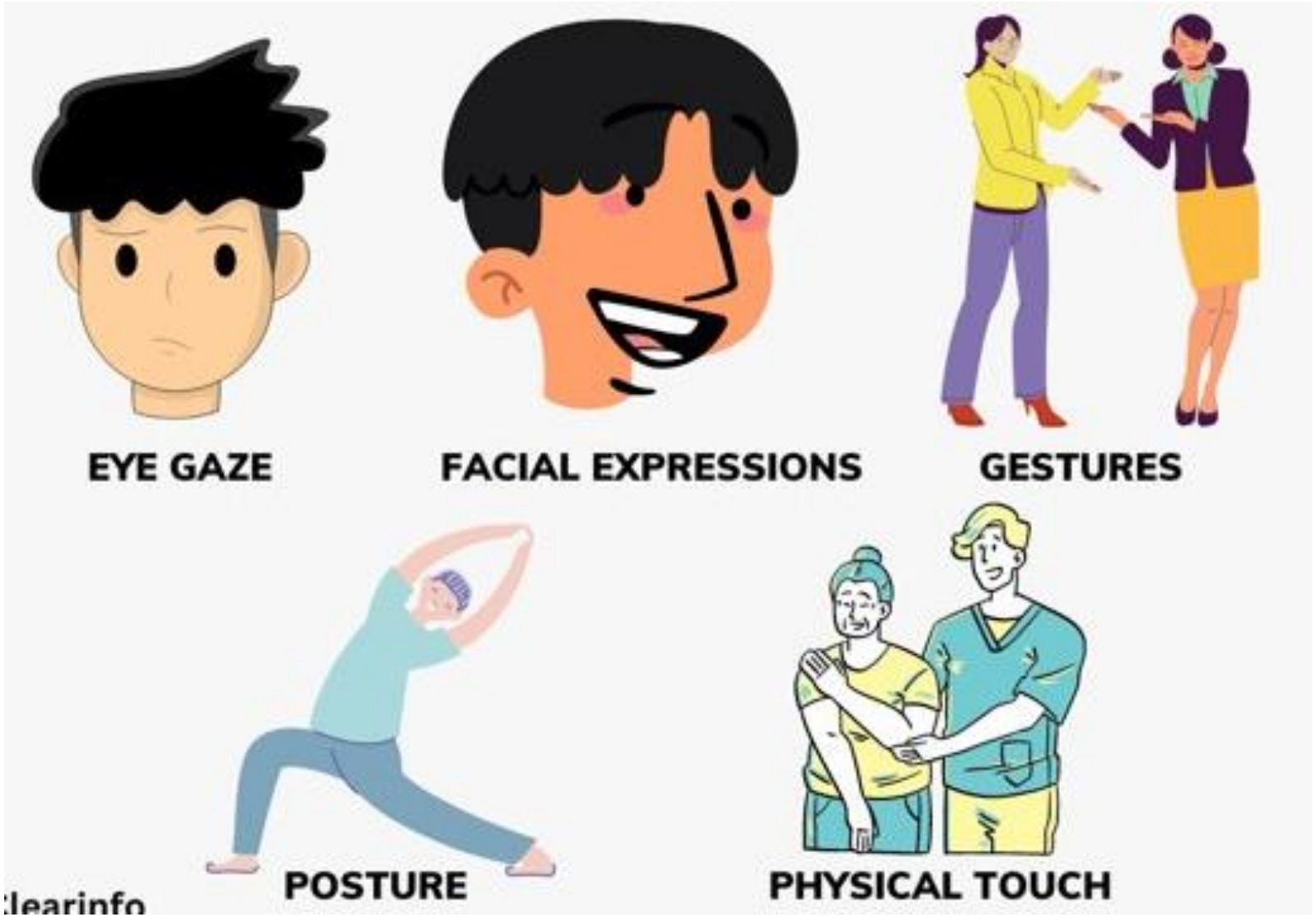
Ask children and their family caregivers, how best to communicate with them!





Family caregivers

- Integral to the pain assessment process
- Reliable and accurate reporters of their child's pain
- Their participation in the assessment of their child's pain allows them to:
  - Draws upon their knowledge regarding the child's communication patterns, mood, and behaviour
  - Facilitates their understanding of their child's cause of and response to pain
  - Provide an opportunity for reflection on ongoing, chronic pain
  - Develops their comfort and confidence with pain assessment and monitoring strategies


EYE GAZE
FACIAL EXPRESSIONS
GESTURES
POSTURE
PHYSICAL TOUCH

Pain history

Pain history

Change in behaviour

Previous pain experiences

Identify any past or current treatments for pain

Change in behavior

- Regular or persistent crying
- Groaning
- Facial grimacing
- Loss of interest in food
- Difficulty sleeping
- Changes in appearance
- Activity level

Past or current treatments for pain

- Includes medications, home remedies and alternative treatments
- Enquire of
  - Timelines
  - Effectiveness
  - Adverse effects

CMC are at increased risk for medication errors

Medical Technology:

Lines/Drains/Airways

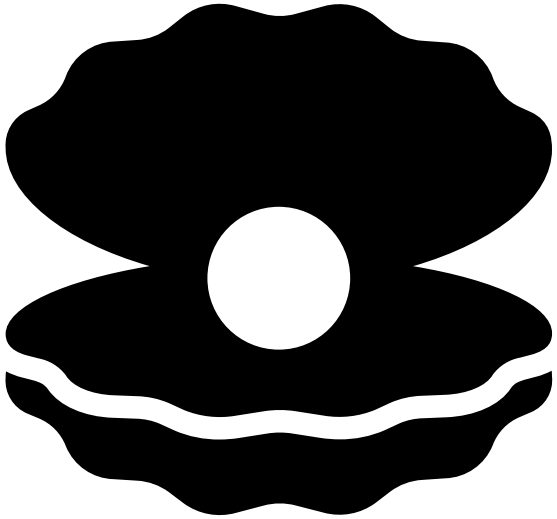
Drain	
Gastrojejunostomy Tube	

Central Venous Catheter	
Port (Single) 02/08/23 Left Subclavian	

Technology reconciliation is also important

Clinical pearl

- When assessing pain in children with SNI, a multidimensional approach includes
  - Assessments of behaviour
  - Communication needs
  - Functional abilities
- Pain assessment tools are available



Non-communicating Children's Pain Checklist – Revised (NCCPC-R)

Non-communicating Children's Pain Checklist – Revised (NCCPC-R)

NAME:_____ UNIT/FILE #: _____ DATE:_____ (dd/mm.yy)

OBSERVER:_____ START TIME:_____AM/PM STOP TIME:_____AM/PM

How often has this child shown these behaviours in the last 2 hours? Please circle a number for each item. If an item does not apply to this child (for example, this child does not eat solid food or cannot reach with his/her hands), then indicate "not applicable" for that item.

0 = NOT AT ALL	1 = JUST A LITTLE	2 = FAIRLY OFTEN	3 = VERY OFTEN	NA = NOT APPLICABLE

I. Vocal

1.	Moaning, whining, whimpering (fairly soft).....	0	1	2	3	NA
2.	Crying (moderately loud).....	0	1	2	3	NA
3.	Screaming/yelling (very loud).....	0	1	2	3	NA
4.	A specific sound or word for pain (e.g., a word, cry or type of laugh).....	0	1	2	3	NA

r-FLACC (revised FLACC) **Pain Rating Scale for children with developmental disability.**

FLACC Scale[2]	0	1	2
1 Face	No particular expression or smile.	Occasional grimace or frown, withdrawn, disinterested.	Frequent to constant frown, clenched jaw, quivering chin.
2 Legs	Normal position or relaxed.	Uneasy, restless, tense.	Kicking, or legs drawn up.
3 Activity	Lying quietly, normal position, moves easily.	Squirming, shifting back and forth, tense.	Arched, rigid or jerking.
4 Cry	No crying (awake or asleep).	Moans or whimpers; occasional complaint.	Crying steadily, screams or sobs, frequent complaints.
5 Consolability	Content, relaxed.	Reassured by occasional touching, hugging or being talked to, distractible.	Difficult to console or comfort.

Activity and Participation

- Review impact on daily life activities:
  - Sleep
  - Eating
  - School
  - Social activities
  - Physical activities
  - Interactions with family and peer interaction

Environment

- Various environments: home, school, extracurricular activities, respite
- Equipment and technology

Personal factors

- Developmental stage
- Sensory profile including vision and hearing impairments
- Language, ethnic, and cultural factors
- Emotional functioning e.g., coping responses to pain
- Presence of psychological symptoms, psychiatric disorders

An approach to pain in child with SNI: Thorough physical examination

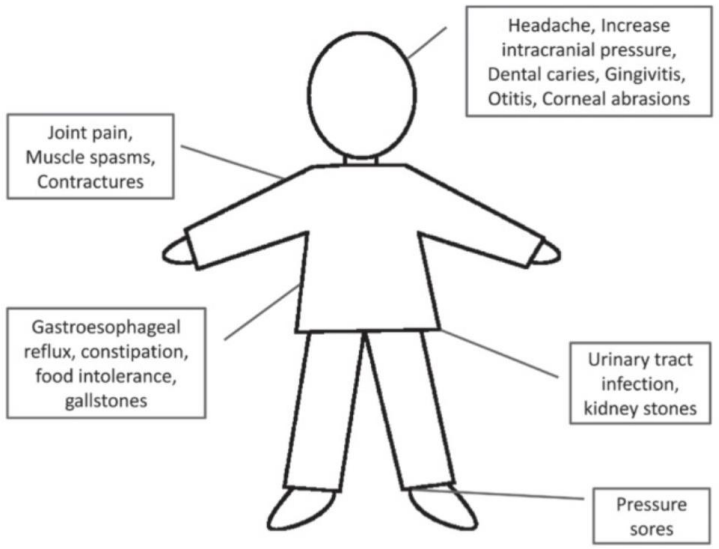
Thorough physical examination

- Vital signs
- Growth parameters
- General observation: posture, gait, cognitive state
- Motor (e.g., increased tone, muscle spasms), functional range of motion of joints
- Autonomic changes (e.g., numbness, hyperesthesia)
- Technology including wheelchair

TABLE 6 Physical Examination as Part of Nociceptive Evaluation

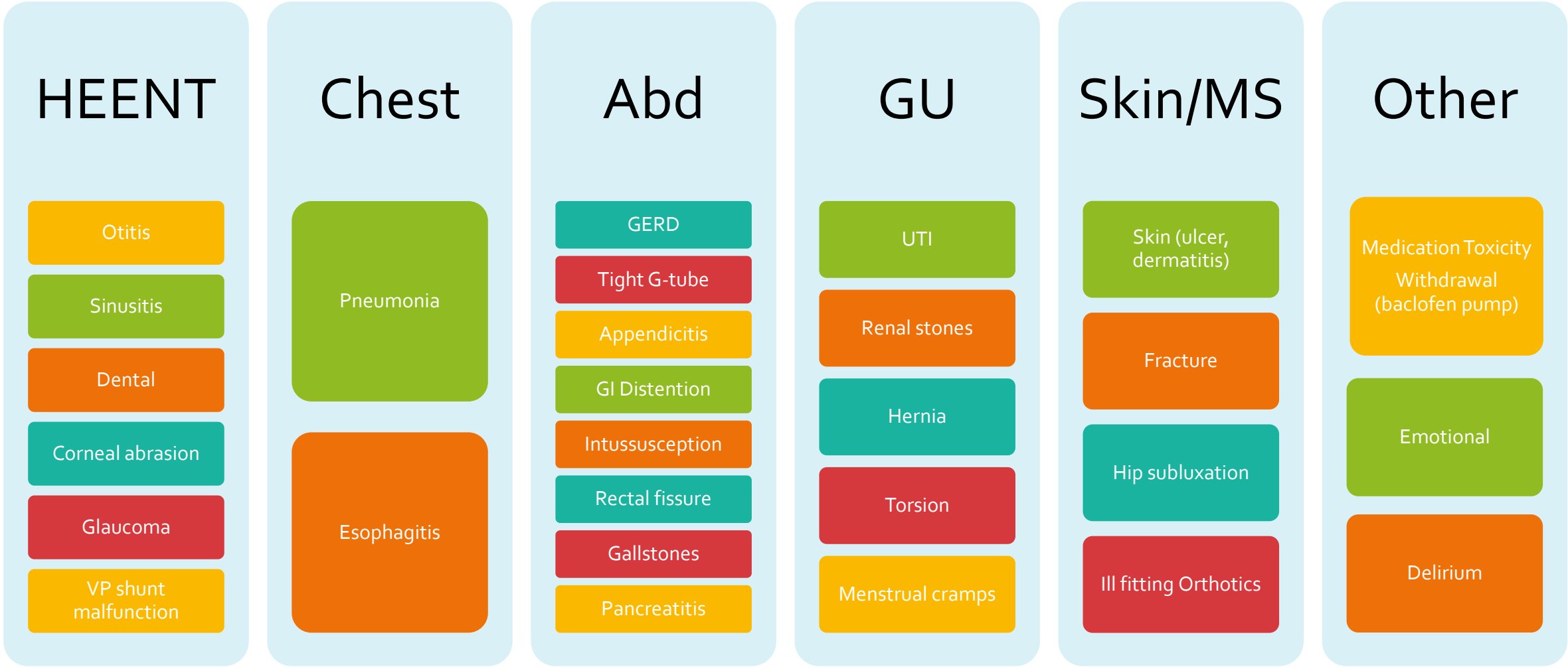
Physical Examination Technique	Potential Nociceptive Pain Source
Inspection of eyes for tears and conjunctival injection	Corneal abrasion
Inspection of dentition, mouth, and throat	Dental caries and abscess, gingivitis, tonsillitis
Inspection and palpation of shunt catheter site	Ventriculoperitoneal shunt malfunction
Inspection and movement of gastrostomy tube	Gastrostomy tube tension associated with growth
Inspection and palpation of abdomen	Constipation, distention
Inspection of skin	Hair tourniquet or pressure ulcer
Inspection, palpation, and movement of extremities	Occult fracture
Palpation and range-of-motion of joints	Subluxation (especially hips)

Hauer J, Houtrow AJ; SECTION ON HOSPICE AND PALLIATIVE MEDICINE, COUNCIL ON CHILDREN WITH DISABILITIES. Pain Assessment and Treatment in Children With Significant Impairment of the Central Nervous System. Pediatrics. 2017 Jun;139(6):e20171002. doi: 10.1542/peds.2017-1002. PMID: 28562301.



Rasmussen & Gregoire, 2015

Acute Pain / Irritability Sources



Hauer and Houtrow 2017; Morse et al 2020; Hauer 2024

Investigations and treatment

- Tailored to clinical assessment
- Often concurrent
- Consider the 3P's of pain management
  - Physical (cold, heat, rest)
  - Psychological (including supporting parents)
  - Pharmacological

Assessment and Trials

- History/exam: VP shunt, ITB pump, G-tube, eyes
- Blood: complete blood count (CBC), metabolic panel (CMP), gamma-glutamyl transferase (GGT), lipase
- Urine: urinalysis and culture
- Stool: *C. difficile* stool testing, fecal calprotectin
- Abdominal imaging: if above negative
- Diagnostic trials: proton pump inhibitor, constipation, dysmenorrhea

Oral Health

- Oral health issues are common and include
  - Dental caries
  - Dental trauma
  - Molar hypomineralisation
  - Drooling
  - Oral hypersensitivity/aversion
  - Oral consequences of tube feeding
- Dental caries are common (25-58%;)

Moursi A, Fernandez J, Daronch M, Zee L, Jones C. Nutritional and Oral Health Considerations in Children with Special Health Care Needs: Implications fo Oral Health Care Providers. *Pediatr Dent*. 2010;32(4).

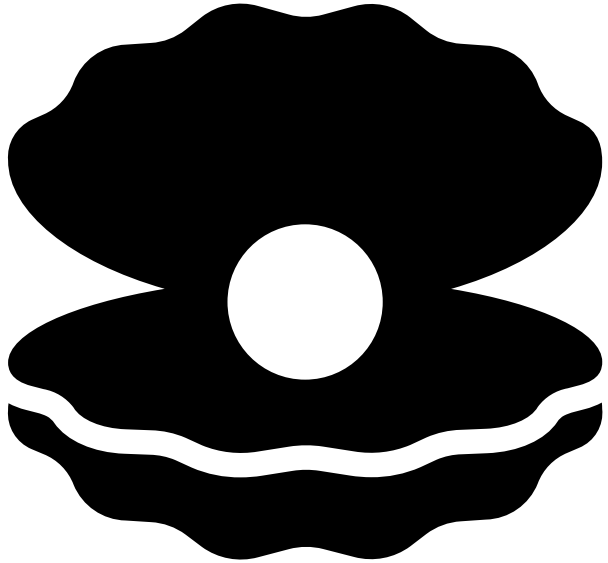
Vozza I. Preventive strategies in oral health for special needs patients. *Ann Stomatol (Roma)*. 2015:96-99. doi:10.11138/ads/2015.6.3.096
Moursi A, Fernandez J, Daronch M, Zee L, Jones C. Nutritional and Oral Health Considerations in Children with Special Health Care Needs: Implications fo Oral Health Care Providers. *Pediatr Dent*. 2010;32(4).

Vozza I. Preventive strategies in oral health for special needs patients. *Ann Stomatol (Roma)*. 2015:96-99. doi:10.11138/ads/2015.6.3.096

Clinical practice pearl

Anticipatory care:
Refer a child with SNI to dentist by their first birthday or within 6 months of the eruption of the first tooth

In a child with SNI presenting in pain:
Consider a dental source of pain



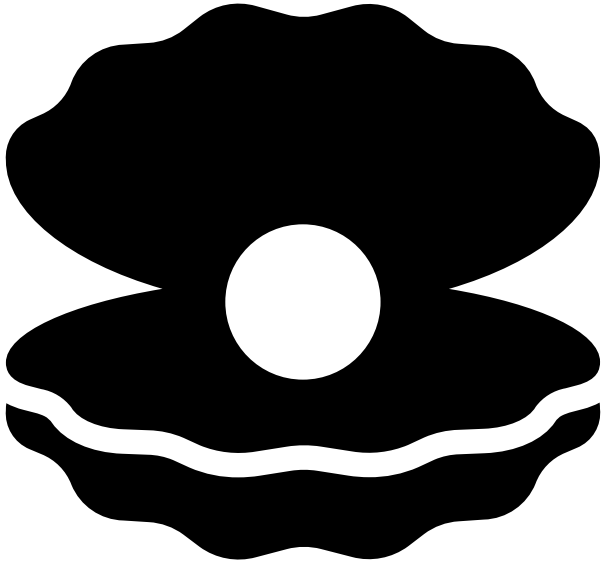
Bone Health

- Multiple risk factors for impaired bone health and bone fragility
- Bone pain can be a presenting symptom of bone fragility
- Impaired bone health including increased fracture risk arises from mechanisms impacting both bone formation and mineralization

Assessment of impaired bone health

- Consider risk factors impacting bone health
- Identify clinical evidence of bone fragility – fractures, bone pain
- Investigations can target:

1. Underlying mechanisms
   - Bone mineralization defect - Calcium, phosphate, creatinine, ALP, PTH and 25 hydroxy-vitamin D
   - Bone formation
2. Quantitation of bone fragility
   - Bone mineral density scan
   - X-rays: lateral spine x-ray vertebral compression fractures.



Clinical practice pearl

Anticipatory care in a child with SNI with no symptoms

Regular review of underlying factors contributing to bone fragility and eliminate or decrease risk factors

Ensure adequate dietary calcium and Vitamin D3 intake – consider starting doses of Vitamin D3 (800-1000 international units)

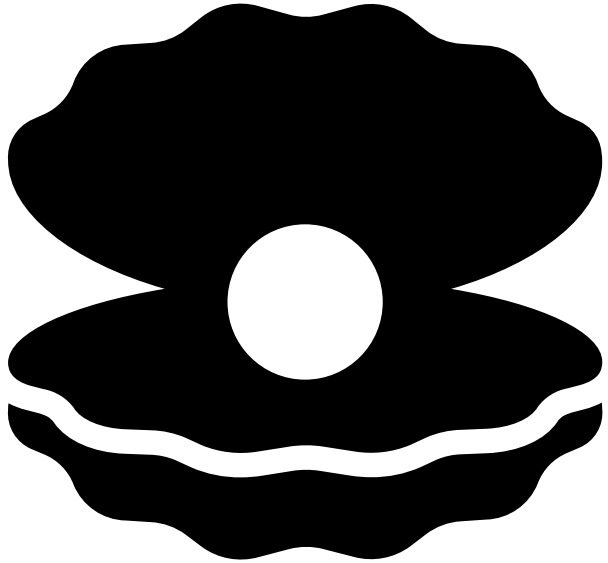
Encourage weight bearing

1-2 yearly lab studies; if abnormality detected – correct deficiency

Clinical practice pearl

Anticipatory care in a child with SNI with symptoms i.e., fractures or bone pain:

- Review of underlying factors contributing to bone fragility and eliminate or decrease risk factors
- Referral to Bone Health Clinic where the following might be considered at time of referral - Laboratory studies (Ca, phosphate, creatinine, ALP, 25(OHD) PTH), Spine X-ray, BMD DXA Scan



Disorders of tone

- Hypertonia: abnormally increased resistance to passive stretch
- Three types of neurologically mediated hypertonia:
  - Spasticity
  - Dystonia:
  - Rigidity

Sanger et al Paeds 2003

Disorders of tone (in reality)

- Hypertonia, hypotonia, spasticity, dystonia, posturing, athetosis, chorea, rigidity, myoclonus

- **Involuntary movement and/or disorders of movement control**

Why treat hypertonia?

- Relieve pain and enhance a child's comfort
- This is particularly true of dystonia
- Ease caregiving
- Occasionally can enhance motor function

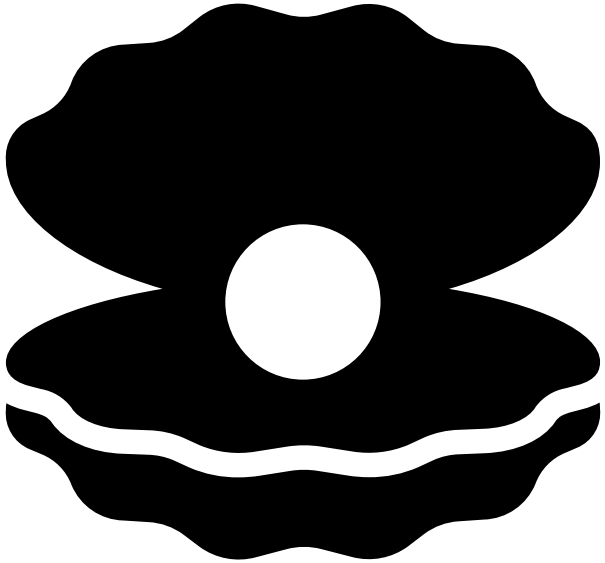
- Variety of treatment guidelines exist (AACPDM, NICE (UK))
  - All emphasize the importance of determining treatment goals at outset

Treatment options

- Stretching
- Medications
  - Baclofen (spasticity, dystonia), Diazepam (spasticity, dystonia), Trihexiphendyl, Dantrolene (spasticity), Gabapentin (spasticity, dystonia), Clobazam (spasticty), Carbidopa (chorea)
- Injections
  - Botulinum toxin
- Implantable devices and surgeries
  - Intrathecal baclofen pump
  - Selective dorsal rhizotomy
  - Deep brain stimulation
  - Multiple level orthopedic surgery (for spasticity?)

However

- Efficacy is variable and unpredictable
- Say that
- Malnutrition, infection, GI distress and sleep deprivation frequently worsen tone.
- If these problems are chronic, consider treating them first



Clinical practice pearl: Stepwise approach

1.What is being experienced?

-Does it impact function, sleep, pain or range of motion?

-Is it a problem? If no. Stop right there. Go out and play!

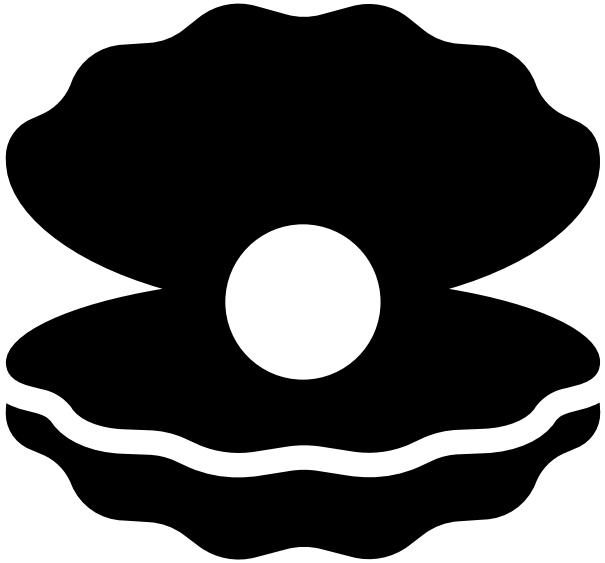
2. What is it?

3. How does it impact life?

4. What makes it better or worse?

5. What has been tried before?

6. What does or what would the person with the problem say?



Clinical practice pearl: Optimize

-Nutrition

-Sleep

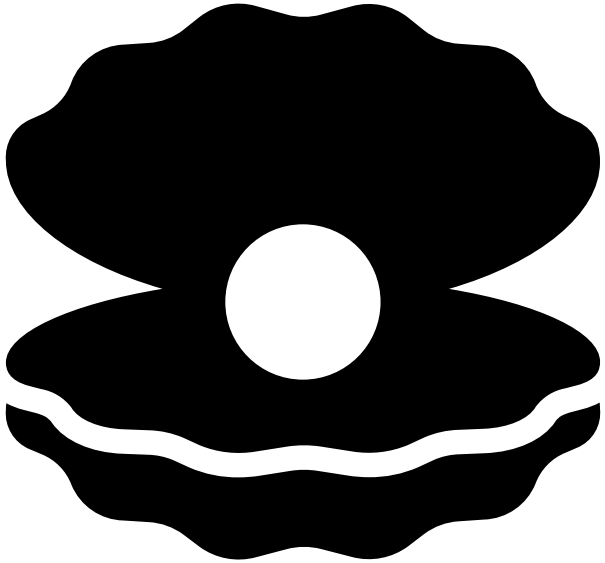
-Pain

-Mood

-Breathing

-Infection

-Bowel and bladder



Clinical practice pearl: Deciding next steps

-“What are your goals?” (short term, long term)

-“If treatment works, what would a minimum **meaningful** change in life look like to you?”

OR

“How will we know that this is successful?”

-“Is it practical to take a drug three times a day?”

-“If this choice is 100% yours, what would you do?”

Stretching

- More than just for range of motion maintenance
- Could reduce spasticity for a period of time - 30-60 minutes?
- Useful prior to activities such as bathing or dressing

Baclofen

- Dosing is quite variable
- Half-life 3-4 hours (three times daily dosing)
- Side effects: sedation, constipation
- Risks: withdrawal, severe
- Cost: low

PM R 10 (2018) 235-244

www.pmrjournal.org

Original Research—CME

Pharmacogenomic Variability of Oral Baclofen Clearance and Clinical Response in Children With Cerebral Palsy

Matthew J. McLaughlin, MD, MS, Yang He, PhD, Janice Brunstrom-Hernandez, MD, Liu Lin Thio, MD, PhD, Bruce C. Carleton, PharmD, Colin J.D. Ross, PhD, MS, Andrea Gaedigk, PhD, Andrew Lewandowski, PhD, Hongying Dai, PhD, William J. Jusko, PhD, J. Steven Leeder, PharmD, PhD

Side effects are real

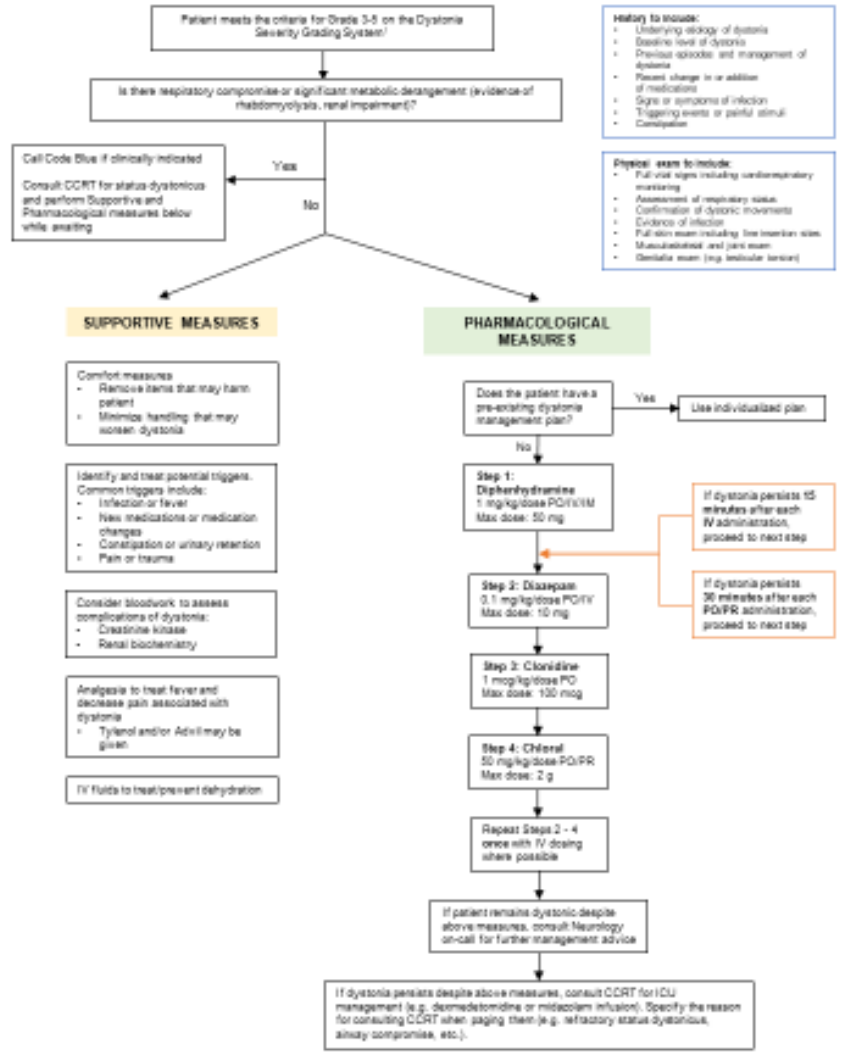
- Range of motion can be a time and energy sink
- Bracing takes time to acquire, adjust, don and doff
- Drugs – sedation, constipation, one more thing to do
- Injections and surgeries are… injections and surgeries!
- Financial Cost

Treatment doesn't work

- Treatment failure is common
- What's the next step?
  - Increase dose?
  - Different drug?
  - Try a different treatment?
  - Do nothing and be ok with that?

Version date: Feb. 2, 2022

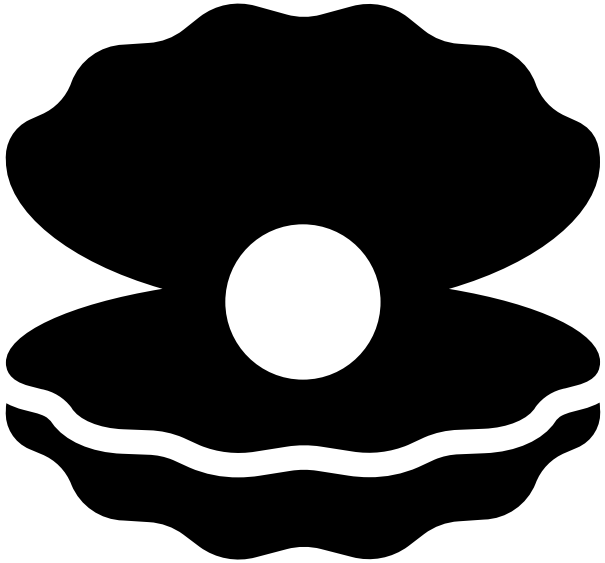
Status Dystonicus Pathway



Hip migration (dysplasia)

- Pain (contractures or osteoarthritis)
- Caregiving challenges
- Seating difficulties
- Decubitus ulcers

- All of the above impact a child's quality of life
- Emphasize the importance of hip surveillance



Clinical practice pearl

Multiple guidelines developed – familiarize with one and use it e.g., AACPDM along with local referral pathways

Typically involves:

- Explaining to family indication for surveillance
- Assessment for pain on history and examination
- Hip abduction on examination
- Radiological examination (AP pelvis; frequency varies by age, clinical situation including GMFCS, exam findings)

Constipation

- Common
- Contributing factors
  - Physiological alterations, e.g., gut dysmotility
  - Medication side effects
  - Lifestyle

Stephens et al. 2017
Veugelers ey a. 2010

Constipation management

1. Disimpaction/ “cleanout”
2. Maintenance
3. Family education

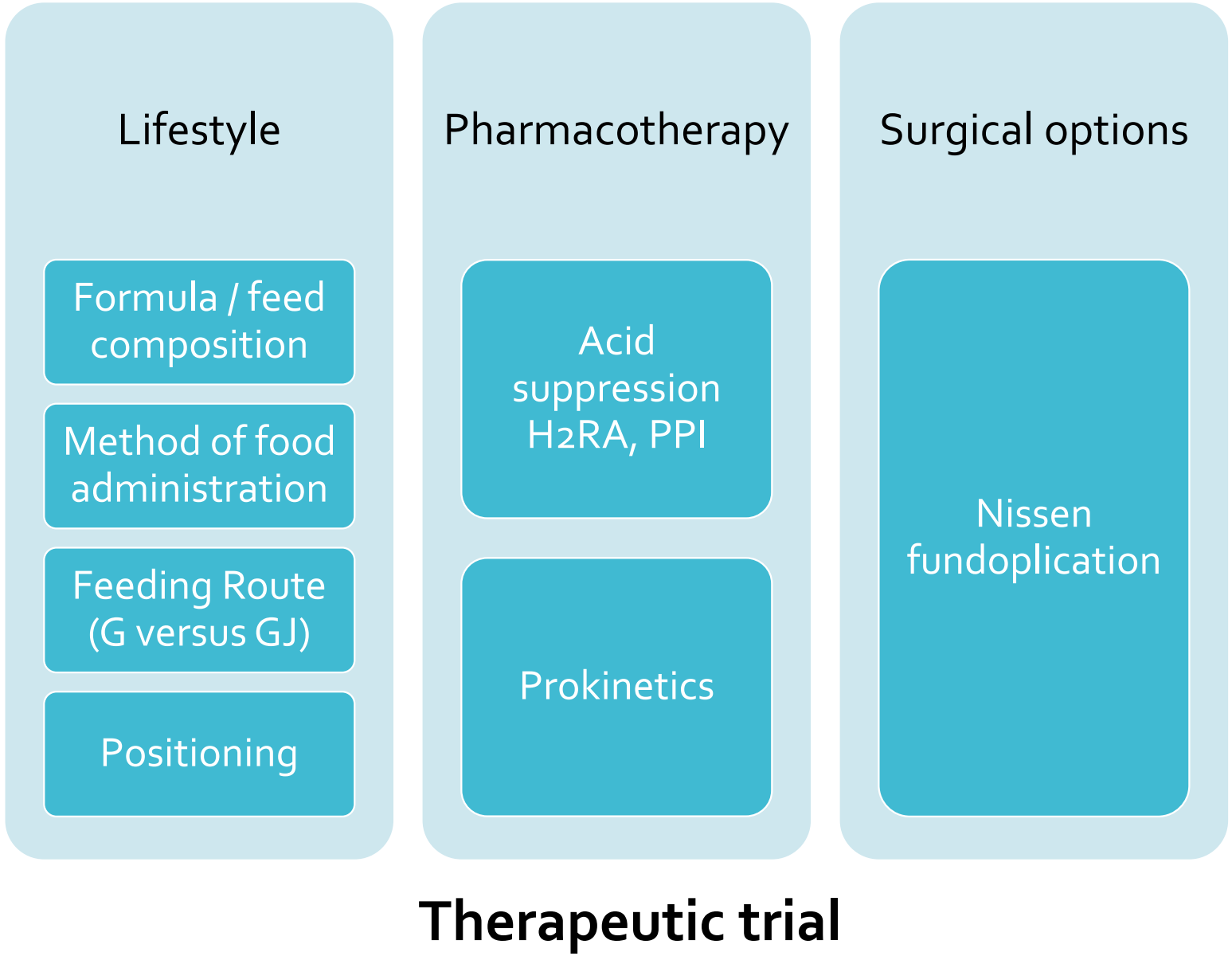
…… be seen as a lower priority than other aspects of medical management

GERD is ALSO common in children with neurologic impairment (upto 70%)

- Contributing factors:
  - Nervous system function
  - Dysmotility
  - Hypotonia
  - Lifestyle

There is a lack of evidence in the treatment of….

Treatment options (GERD)



Therapeutic trial

A reminder to me

People with disabilities experience pain and general discomfort for the same reasons I do – headache, dysmenorrhea, thirst, hunger, psychological or emotional distress.

Chronic pain in children with SNI

Acute versus chronic pain

ACUTE

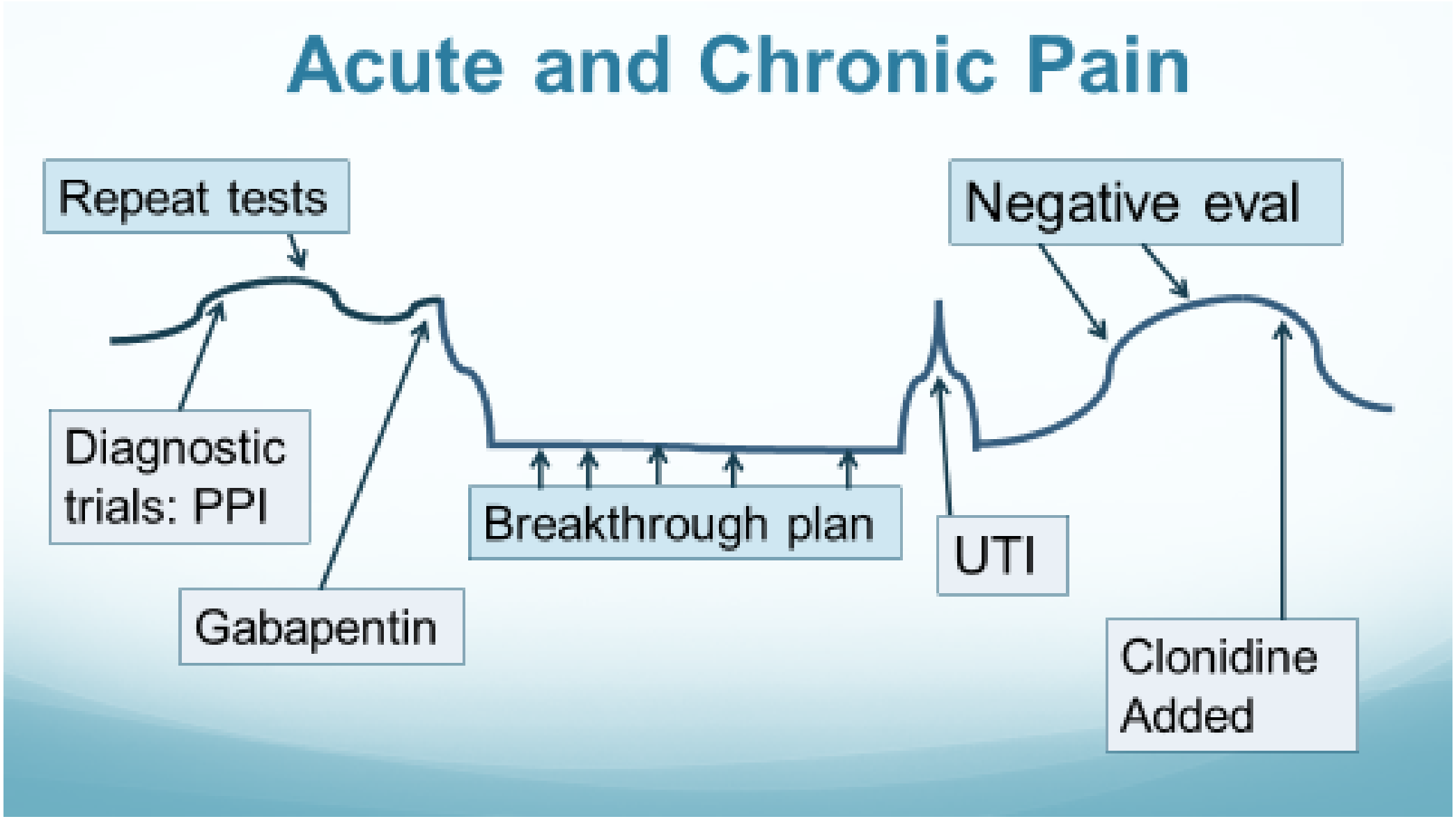
- Caused by tissue inflammation or injury (nociceptive)
- Hours – days
- Diagnostic tests can be helpful
- Resolution expected after treatment and healing

CHRONIC

- Variety of categories – musculoskeletal, neuropathic, visceral
- Persistent; > 3months by definition
- Diagnostic tests often unhelpful
- Focus is often on pain control

Hauer J, Houtrow AJ; SECTION ON HOSPICE AND PALLIATIVE MEDICINE, COUNCIL ON CHILDREN WITH DISABILITIES. Pain Assessment and Treatment in Children With Significant Impairment of the Central Nervous System. Pediatrics. 2017 Jun;139(6):e20171002. doi: 10.1542/peds.2017-1002. PMID: 28562301.

Acute and Chronic Pain



Acute and Chronic Pain / Irritability

Nociceptive and Other	Neuro-problems	Neuro-pain*
• Fracture • Cornea • Musculo-skeletal • Drug toxicity	• Spasticity • Dystonia • Seizures • Dementia	• Central • Visceral • Autonomic

***NEURO-PAIN**: chronic pain sources due to altered nervous system in children with SNI without diagnostic tests or distinct features to differentiate one from another, with similar chronic treatment strategies

American Academy of Pediatrics
DEDICATED TO THE HEALTH OF ALL CHILDREN™

Pain Assessment and Treatment in Children With Significant Impairment of the Central Nervous System

Julie Hauer, MD, FAAP,[a,b] Amy J. Houtrow, MD, PhD, MPH, FAAP,[c]
SECTION ON HOSPICE AND PALLIATIVE MEDICINE, COUNCIL ON CHILDREN WITH DISABILITIES

Clinical Report – Reaffirmed With Reference & Data Updates

This Clinical Report has been reaffirmed with reference and data updates. New or updated references and datapoints are indicated in bold typeface. No other changes have been made to the text or content.

The AAP would like to acknowledge Julie Hauer, MD, FAAP, FAAHPM, and Amy Houtrow, MD, PhD, MPH, FAAP, for these updates.

abstract

FREE

[a]Complex Care Service, Division of General Pediatrics, Boston Children's Hospital, Assistant Professor, Harvard Medical School, Boston Massachusetts; [b]Seven Hills Pediatric Center, Groton, Massachusetts; and [c]Department of Physical Medicine and Rehabilitation, University of Pittsburgh, Pediatric Rehabilitation Medicine, Rehabilitation Institute, Children's Hospital of Pittsburgh of UPMC, Pittsburgh, Pennsylvania

Non-pharmacologic strategies

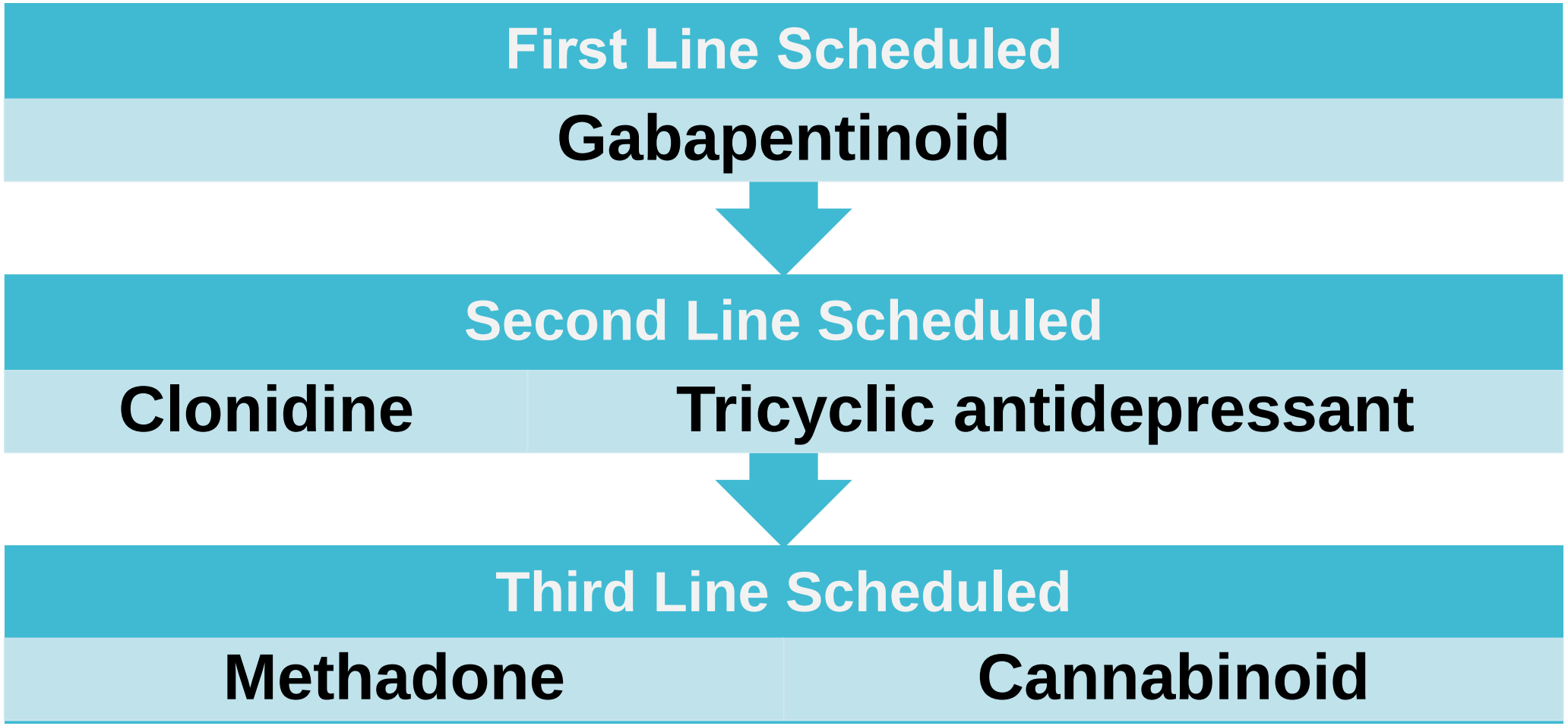
- **Scheduled** (benefit for neuro-problems and neuro-pain)
  - Music therapy
  - Massage therapy / Deep pressure / Vibration
  - Pool therapy / Whirlpool
  - Accupressure, Accupuncture
- **Breakthrough strategies**
  - See AAP clinical report 2017 for details
  - PT, OT, Child Life

Hauer and Houtrow 2017, Yuanjie Y, et al. Acupuncture in the Treatment of Abnormal Muscle Tone in Cerebral Palsy

Suggestions

- Scheduled medication
- Manage co-morbid problems:
  - sleep, spasticity, dystonia, seizures, GERD
- Acute on Chronic pain
- Triggers:
  - Environment
  - GI tract distention

Medication Trials



As needed: clonidine, opioid, benzo

Gastrointestinal (GI) tract distention

- **Neuro-pain lowers GI tract pain threshold**
- **Constipation**
- **Excessive intestinal gas**
  - Drugs: polyethylene glycol, lactulose, PPI
  - Small intestinal bacterial overgrowth (SIBO)
  - Vent G-tube
- **Excess calories or osmolality of feeds**

Hauer and Houtrow 2017; Hauer 2024

Calorie increases

- **Vicious cycle**
  - Unrecognized chronic pain
  - Pain increases metabolism
  - Calories are increased
- **Break the cycle**
  - Treat chronic pain
  - Put aside weight gain for 3 months
  - 25-30% calorie decrease in context of weight goal

Gale R, et al 2016, Hauer 2017, Hauer 2023

Social Worker

"I think pain without a source is pernicious - it steals comfort and ease from the child; it creates suffering and fear for the parents. Parents want to comfort their child - pain and irritability prevent this human need. Solace is impossible. Parents have been driven to the brink of despair; Neighbours have called child welfare (as they do not understand why the child keeps crying). I cannot think of another condition in pediatric medicine that causes such pernicious distress."

Family Caregiver

"It is not easy to answer - treating pain made us feel normal again …gave us more of a normal life. At the time …I wished I wasn't a Mom any more …Now all I want to be is her Mom. It was hard!"

Partnership with parents, caregivers and other professionals

Hauer
and Soldiuk 2017

Name and Frame

Partnership with Parents

Fear, Risk tolerance, Shared decision making

Information need *versus* Emotional need

"You need to do more tests to figure this out"

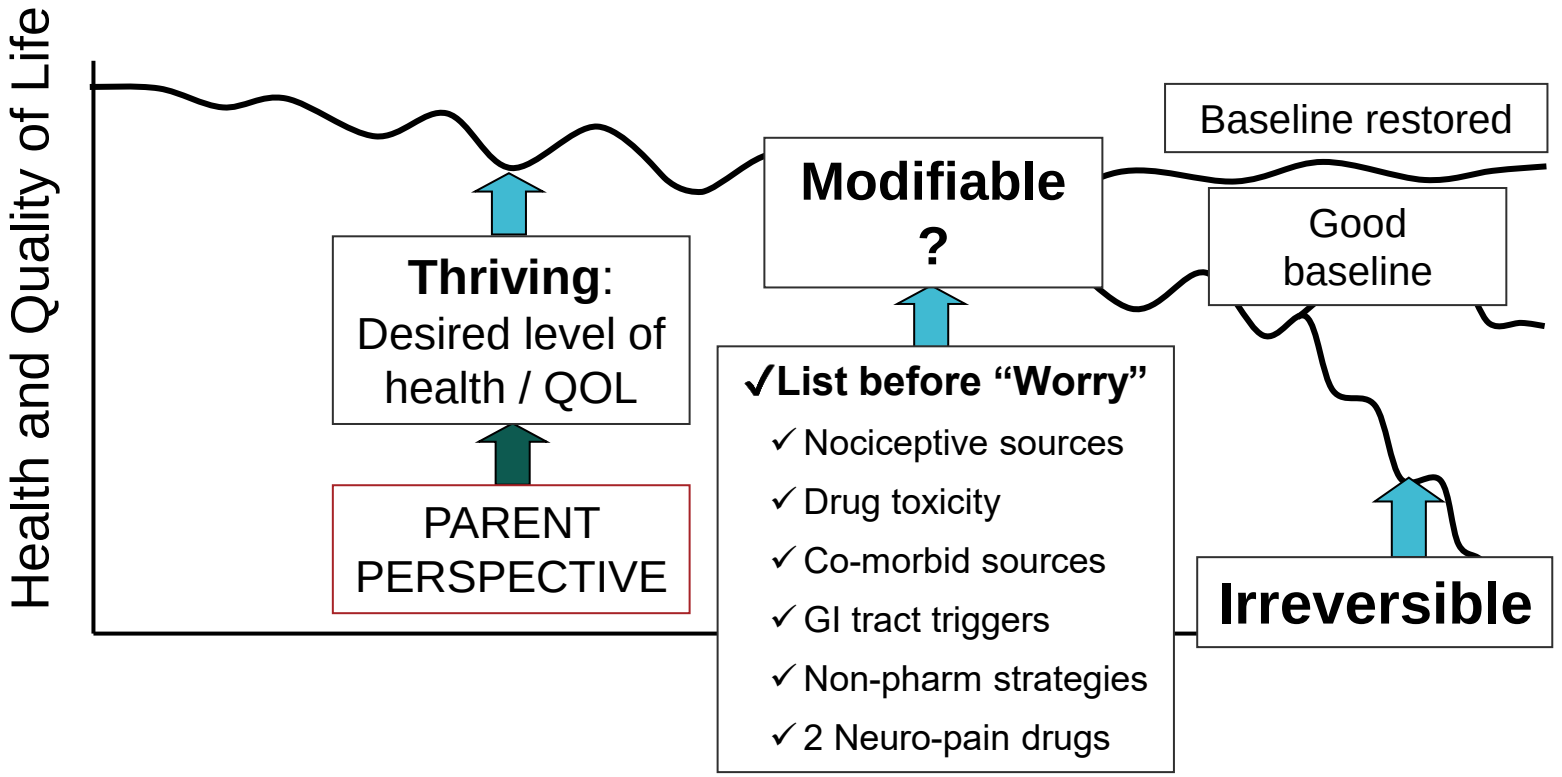
- **Information** We have done many tests…
- **Emotion** It must be hard to hear… tell me more…

What if a problem is missed / delayed?

Iterative Naming & Framing Statements

Hauer 2024

Anticipatory statement (adjust to parent)	Framework
I expect benefit with this 1st / 2nd drug (intervention) though I can't guarantee this. Many children like have benefit, not all have the same degree of benefit.	**Goal achieved or "Baseline restored"**
Benefit is possible with this 3rd intervention though it may not be as much as we want or have seen.	**"Good baseline" possible**
I am worried that X's problems are becoming greater than our best treatments. I will share why I am concerned. Your view is equally important.	**"Irreversible" possible**



Chronic Problems Without Cure

Chronic pain due to the impaired CNS

Symptom Escalation During Decline

- **“Less is More”**
  - Hold mucous mobilizing interventions
  - Decrease feeds/fluids
  - Hold nonessential medications
- A new baseline may occur
- Adjust as needed

Case Discussion

You are a physician in an outpatient clinic. A patient you follow, Jonathan, a 4-year-old boy with cerebral palsy, GFMCS V, presents to your clinic for routine follow up. Apart from the cerebral palsy, Jonathan has a history of (i) seizures, well controlled on Keppra; (ii) GERD, on lansoprazole; (iii) constipation, on PEG3350; (iv) hypertonia; and (v) mild scoliosis).

Jonathan's mother's main concern is that he appears more uncomfortable, often wincing and crying. He is having difficulty sleeping now as well, and his physiotherapy sessions are often cut short due to him appearing in pain. She doesn't know why this has been happening, but it has been ongoing for over 4 months now, maybe worsening slightly over that time. The mother reports it is also affecting her sleep and impacting her productivity at work.

Support for Families

What can we do?

- Acknowledge our own biases and lived experiences
- Empower parents as the expert in their child
  - Validate
  - Highlight strengths
- Consistency and communication
  - Parent and child
- Explore the ways in which the child communicates

Coping with Uncertainty

- Acknowledge that there may not be a certainty for the source of pain
- Explore how the family view and defines suffering
- Psychoeducation to offset blaming, feelings of inadequacy, and helplessness
- Exploration of environmental or social interventions at home that they can control

References (i)

Lundy C, Doherty G, Fairhurst C. Botulism toxin type A injections can be an effective treatment for pain in children with hip spasms and cerebral palsy. *Dev Med Child Neurol.* 2009;51:705e710

Smith H, Elliott J. Alpha(2) receptors and agonists in pain management. *Curr Opin Anaesthesiol*. 2001;14(5):513–518.

Liow NY, Gimeno H, Lumsden DE, Marianczak J, Kaminska M, Tomlin S, Lin JP. Gabapentin can significantly improve dystonia severity and quality of life in children. *Eur J Paediatr Neurol.* 2016 Jan;20(1):100-7.

Bonouvrié LA, van Schie PE, Becher JG, van Ouwerkerk WJ, Vermeulen RJ. Intrathecal baclofen for progressive neurological disease in childhood: a systematic review of literature. *Eur J Paediatr Neurol.* 2012 May;16(3):279-84.

Kraus T, Gegenleitner K, Svehlik M, Novak M, Steinwender G, Singer G. Long-term therapy with intrathecal baclofen improves quality of life in children with severe spastic cerebral palsy. *Eur J Paediatr Neurol.* 2017 May;21(3):565-569.

Roubertie A, Mariani LL, Fernandez- Alvarez E, Doummar D, Roze E. Treatment for dystonia in childhood. *Eur J Neurol*. 2012;19(10):1292–1299.

Gormley ME Jr, Krach LE, Piccini L. Spasticity management in the child with spastic quadriplegia. *Eur J Neurol.* 2001 Nov;8 Suppl 5:127-35.

Delgado MR, Hirtz D, Aisen M, et al; Quality Standards Subcommittee of the American Academy of Neurology; Practice Committee of the Child Neurology Society. Practice parameter: pharmacologic treatment of spasticity in children and adolescents with cerebral palsy (an evidence-based review): report of the Quality Standards Subcommittee of the American Academy of Neurology and the Practice Committee of the Child Neurology Society. *Neurology*. 2010;74(4):336–343.

Baguley IJ, Cameron ID, Green AM, Slewa-Younan S, Marosszeky JE, Gurka JA. Pharmacological management of dysautonomia following traumatic brain injury. *Brain Inj*. 2004;18(5):409–417.
34. Smith H, Elliott J. Alpha(2) receptors and agonists in pain management. *Curr Opin Anaesthesiol*. 2001;14(5):513–518.

Hauer J. Feeding Intolerance in Children with Severe Impairment of the Central Nervous System: Strategies for Treatment and Prevention. *Children (Basel).* 2017 Dec 22;5(1).

Hauer J. Identifying and managing sources of pain and distress in children with neurological impairment. *Pediatr Ann.* 2010;39:198-205.

Collins A, Mannion R, Broderick A, Hussey S, Devins M, Bourke B. Gabapentin for the treatment of pain manifestations in children with severe neurological impairment: a single-centre retrospective review. *BMJ Paediatr Open.* 2019 Jul 21;3(1):e000467.

References (ii)

Hauer JM, et al. Gabapentin successfully manages chronic unexplained irritability in children with severe neurologic impairment. *Pediatrics.* 2007;119(2):e519–e522.

Finnerup NB, Otto M, McQuay HJ, Jensen TS, Sindrup SH. Algorithm for neuropathic pain treatment: an evidence based proposal. *Pain*. 2005;118(3):289–305 .

Dworkin RH, O'Connor AB, Backonja M, et al. Pharmacologic management of neuropathic pain: evidence-based recommendations. *Pain*. 2007;132(3):237–251.

Moulin D, Boulanger A, Clark AJ, et al; Canadian Pain Society. Pharmacological management of chronic neuropathic pain: revised consensus statement from the Canadian Pain Society. *Pain Res Manag*. 2014;19(6):328–335.

Stallard P, Williams L, Velleman R, Lenton S, McGrath PJ, Taylor G. PICIC (Pain indicator for communicatively impaired children). *Pain.* 2002 Jul;98(1-2):145-9.

Seidel S, Aigner M, Ossege M, Pernicka E, Wildner B, Sycha T. Antipsychotics for acute and chronic pain in adults. *Cochrane Database Syst Rev*. 2013;(8):CD004844.

Zernikow B, Michel E, Craig F, Anderson BJ. Pediatric palliative care: use of opioids for the management of pain. *Paediatr Drugs*. 2009;11(2):129–151.

Roubertie A, Mariani LL, Fernandez- Alvarez E, Doummar D, Roze E. Treatment for dystonia in childhood. *Eur J Neurol*. 2012;19(10):1292–1299.

Hauer JM. Caring for Children Who Have Severe Neurological Impairment. Baltimore, MD: Johns Hopkins University Press; 2013.

Harrop EJ, Brombley K, Boyce K. Fifteen minute consultation: Practical pain management in paediatric palliative care. *Arch Dis Child Educ Pract Ed.* 2017;102:239–243.

Quinn BL, Solodiuk JC, Morrill D, Mauskar S. CE: Original Research: Pain in Nonverbal Children with Medical Complexity: A Two-Year Retrospective Study. *Am J Nurs.* 2018 Aug;118(8):28-37.

Bajelidze G, Belthur MV, Littleton AG, Dabney KW, Miller F. Diagnostic evaluation using whole-body technetium bone scan in children with cerebral palsy and pain. *J Pediatr Orthop.* 2008 Jan-Feb;28(1):112-7.

Ramstad K, Jahnsen RB, Terjesen T. Severe hip displacement reduces health-related quality of life in children with cerebral palsy. *Acta Orthop.* 2017 Apr;88(2):205-210.

Krekmanova L, Hakeberg M, Robertson A, Braathen G, Klingberg G. Perceived oral discomfort and pain in children and adolescents with intellectual or physical disabilities as reported by their legal guardians. *Eur Arch Paediatr Dent.* 2016 Aug;17(4):223-30.

Gössler A, Schalamon J, Huber-Zeyringer A, Höllwarth ME. Gastroesophageal reflux and behavior in neurologically impaired children. *J Pediatr Surg.* 2007;42:1486–1490.

Drooling:
Assessment and Management for Children with Medical Complexity

Learning Objectives

- To describes the types of sialorrhea and common etiologies
- To assess and evaluate the severity and impact of sialorrhea
- To understand common sialorrhea treatment options

Outline

- Case Presentation
- Etiology
- Assessment
  - Physical Exam
  - Tools
- Management Options
  - Observation
  - Rehabilitation
  - Pharmacologic
  - Surgical

Case Presentation

3-year-old boy presenting for routine follow up. Mother notes significant drooling.

PMHx:

- Cerebral Palsy GMFCS V
- Hypertonia
- Seizures disorder
- Suspected asthma
- Scoliosis
- Constipation
- Fed via G-Tube

Medications

- Baclofen
- Keppra
- Clobazam
- PEG-3370
- Ventolin PRN

Definitions

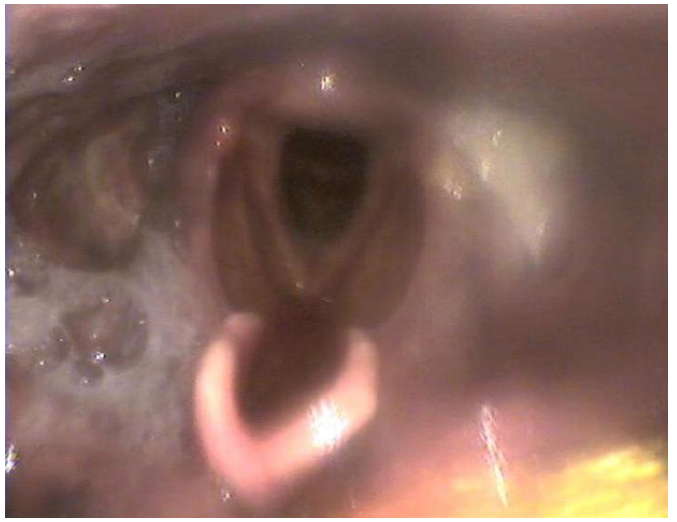
Hypersalivation[1]

Drooling[1]

Sialorrhea[1]

Drooling

Anterior Drooling[2]

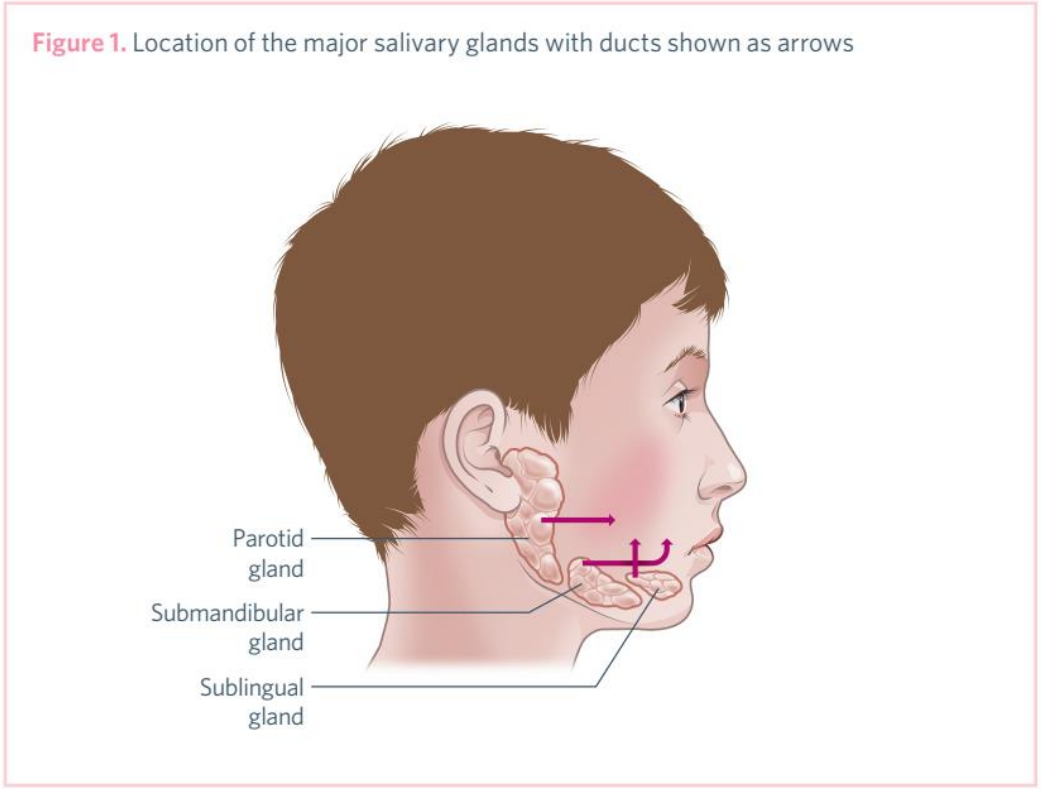


Posterior Drooling[2]

Saliva

- Three major pairs of salivary glands[3]
  - Submandibular gland
  - Parotid gland
  - Sublingual gland

- Functions of saliva[3]
  - Lubricates food, tongue, lips
  - Oral hygiene
  - Bacteriostatic/ bactericidal effects
  - Initiates carbohydrate digestion
  - Regulated esophageal acidity



Sequence of Swallowing[5]

Oral preparatory (voluntary)

Oral (voluntary)

Pharyngeal (involuntary)

Esophageal (involuntary)

Drooling

Overproduction of saliva[1]

Inefficient control of salivary secretions[1]

- Sensory issues
- Motor issues

Etiology[6]

Increase saliva production

- Anticonvulsants (clobazam/ clonazepam)
- Antipsychotic medications
- Toxicity (selenium, mercury…)

Nasal blockage (mouth breathing)

- Adenoid/tonsillar hypertrophy
- Allergic rhinitis

Oral cavity

- Dental malocclusion
- Poor lip closure
- Caries, gum disease, ulcers

Etiology[6]

Inefficient or deficient swallowing

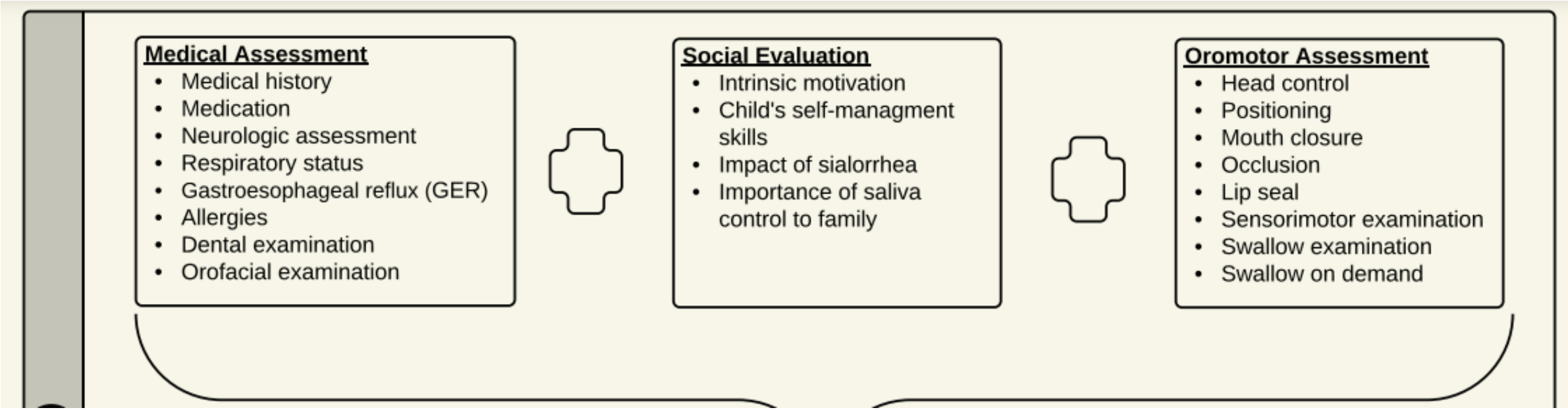
- Anatomic
- Neurologic
  - Cerebral Palsy
  - Bell's Palsy
- Developmental

Hypotonia, poor head control, poor posture

Neurodevelopmental concerns

- Autism
  Severe cognition/ awareness difficulties (sensory deficiency)

Assessment[4]



Impact of Drooling

Clinical Implications[5]

- Aspiration pneumonia
- Perioral dermatological issues
- Dentition problems

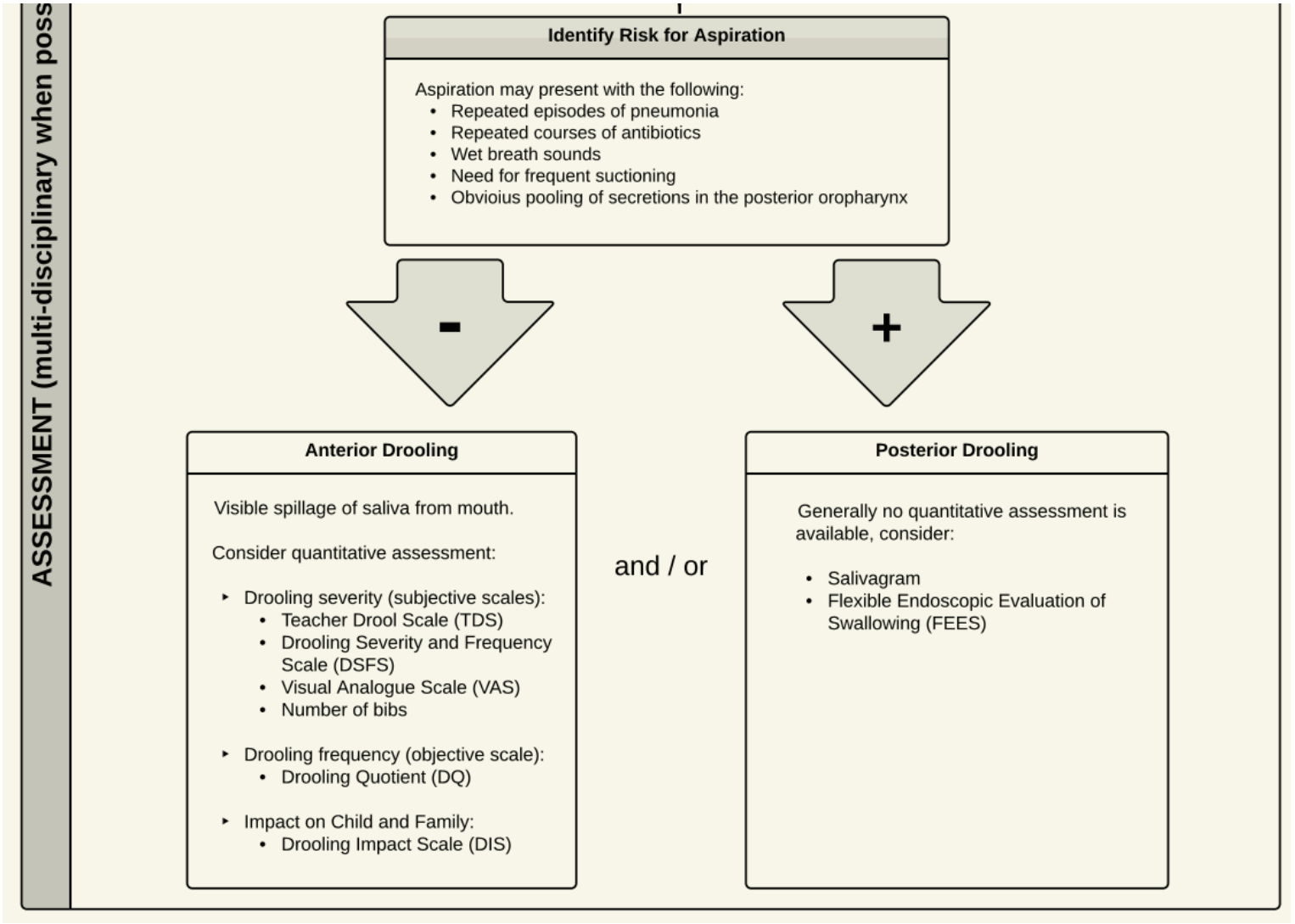
Social Implications[5]

- Impact on self-esteem
- Unpleasant odor
- Embarrassment
- Reduced social inclusion

Focused Physical Exam[7]

- Head and body posture
- HEENT exam
  - Oral cavity
  - Facial exam
  - Tonsils size
- Neurological evaluation
- Pulmonary and cardiac auscultation
- Nutritional status

Assessment[4]



Assessment

- No universally accepted measurement tool[7]
- Few pediatricians use standardised methods to measure[7]
  - Sialorrhea
  - Effectiveness of medications
  - Adverse effects
- Most commonly used measures
  - Count the number of bib changes per day (80%)[7]
  - Degree of parental satisfaction (85%)[7]

Do you use a formal assessment tool when diagnosis or monitoring sialorrhea?

Assessment Tools[7]

Table 1 Measures of sialorrhea

Type of measure	Name of measure
Quantitative/semiquantitative outcome methods	Bib count [20] Bib weight [21] Sochaniwskyj's technique [22] Drooling Quotient [23] 5-min Drooling Quotient (DQ5) [24]
Scales and questionnaires measuring severity	Drooling Infants and Preschoolers Scale (DRIPS) [25] Drooling Severity and Frequency Scale (DSFS) [26] Blasco Index for the assessment of drooling [1] Teacher Drool Scale (TDS) [27] Modified Teacher Drool Scale (mTDS) [28] Visual Analogue Scale (VAS) [29]
Scales and questionnaires measuring severity, impact on quality of life and daily life	Modified drooling questionnaire [30] Drooling Impact Scale (DIS) [31] French version of Drooling Impact Scale (DIS-F) [32] Brazilian Portuguese language version of DIS [33] Drooling impact questionnaire (short version) [6] Questionnaire to evaluate impact of drooling on daily living (questionnaire 1; questionnaire 2) [8] Daniel Drooling Impact Score Questionnaire (DDISQ) [34] Drool rating scale [35]

Assessment

5-Minute Drooling Quotient (DQ5)[9]

- Assesses severity of drooling
- Observed during two trials of 5 minutes
  - Activity
  - Rest

DQ5 Activity

Interaction/singing/playing

	Observation intervals 15 sec	Score 1 of 0
1	0.00-0.15	
2	0.15-0.30	
3	0.30-0.45	
4	0.45-1.00	
5	1.00-1.15	
6	1.15-1.30	
7	1.30-1.45	
8	1.45-2.00	
9	2.00-2.15	
10	2.15-2.30	
11	2.30-2.45	
12	2.45-3.00	
13	3.00-3.15	
14	3.15-3.30	
15	3.30-3.45	
16	3.45-4.00	
17	4.00-4.15	
18	4.15-4.30	
19	4.30-4.45	
20	4.45-5.00	

DQ5 Rest

Watching television/listening to music

	Observation Intervals 15 sec	Score 1 of 0
1	0.00-0.15	
2	0.15-0.30	
3	0.30-0.45	
4	0.45-1.00	
5	1.00-1.15	
6	1.15-1.30	
7	1.30-1.45	
8	1.45-2.00	
9	2.00-2.15	
10	2.15-2.30	
11	2.30-2.45	
12	2.45-3.00	
13	3.00-3.15	
14	3.15-3.30	
15	3.30-3.45	
16	3.45-4.00	
17	4.00-4.15	
18	4.15-4.30	
19	4.30-4.45	
20	4.45-5.00	

Score: $\frac{\text{Total amount of intervals with new saliva}}{20} \times 100 =$

Assessment Tools

Drooling Impact Scale[8]

- Measures impact of drooling
- Evaluates longitudinal changes

THE DROOLING IMPACT SCALE

Over the past week:

1. **How frequently did your child dribble?**

   Not at all 1 2 3 4 5 6 7 8 9 10 Constantly

2. **How severe was the drooling?**

   Remained dry 1 2 3 4 5 6 7 8 9 10 Profuse

3. **How many times a day did you have to change bibs or clothing due to drooling?**

   Once or not at all 1 2 3 4 5 6 7 8 9 10 10 or more

4. **How offensive was the smell of the saliva on your child?**

   Not offensive 1 2 3 4 5 6 7 8 9 10 Very offensive

5. **How much skin irritation has your child had due to drooling?**

   None 1 2 3 4 5 6 7 8 9 10 Severe rash

6. **How frequently did your child's mouth need wiping?**

   Not at all 1 2 3 4 5 6 7 8 9 10 All the time

7. **How embarrassed did your child seem to be about his/her dribbling?**

   Not at all 1 2 3 4 5 6 7 8 9 10 Very embarrassed

8. **How much do you have to wipe or clean saliva from household items eg toys, furniture, computers etc?**

   Not at all 1 2 3 4 5 6 7 8 9 10 All the time

9. **To what extent did your child's drooling affect his or her life?**

   Not at all 1 2 3 4 5 6 7 8 9 10 Greatly

10. **To what extent did your child's dribbling affect you and your family's life?**

    Not at all 1 2 3 4 5 6 7 8 9 10 Greatly

Assessment[10]

Table 1. Thomas-Stonell and Greenberg scale[8]

Drooling severity score	Drooling frequency score
Dry	1) Never
Mild-wet lips	2) Occasionally
Moderate-wet lips and chin	3) Frequently
Severe-drool extends to clothes wet	4) Constantly
Profuse-clothing, hands and objects wet	

Assessment Tools

- Tools to quantify drooling[8]
  - Drooling Impact Scale
  - Drooling Quotient
  - Drooling Severity and Frequency Scale
  - Daniel Drooling Impact Score Questionnaire – DDISQ
  - Modified Teacher's Drooling Scale
  - Weighing oral cotton rolls or bibs
  - Visual Analogue Scale

Case

- Drooling since an infant, but now worsening.
- He is coughing every day now and has noisy breathing when he lies down
- Wiping saliva off his face all day, because of this, kids play with him less
- Doing less physiotherapy, as having to spend time cleaning drool throughout sessions
- Parents anxious+++ about drooling, impacting their activities and sleep as well

What therapies can we offer?

Treatment Options[11]

- Observation
- Rehabilitative / Non-Pharmacologic Measures
- Pharmacologic Measures
- Botulinum Toxin Injection
- Surgical

Consider Alternative Diagnoses

Observations

1. When symptoms were mild to moderate
2. <4 years old with:
    - Anticipation of normal swallowing development and
    - Without pulmonary complications
3. Minimal impact on the quality of life
4. Not a concern for the caregivers

Rehabilitative Options

1. Positioning and posture adjustments
2. Oral motor and oral sensory therapy
3. Behavior therapy
4. Oral appliances

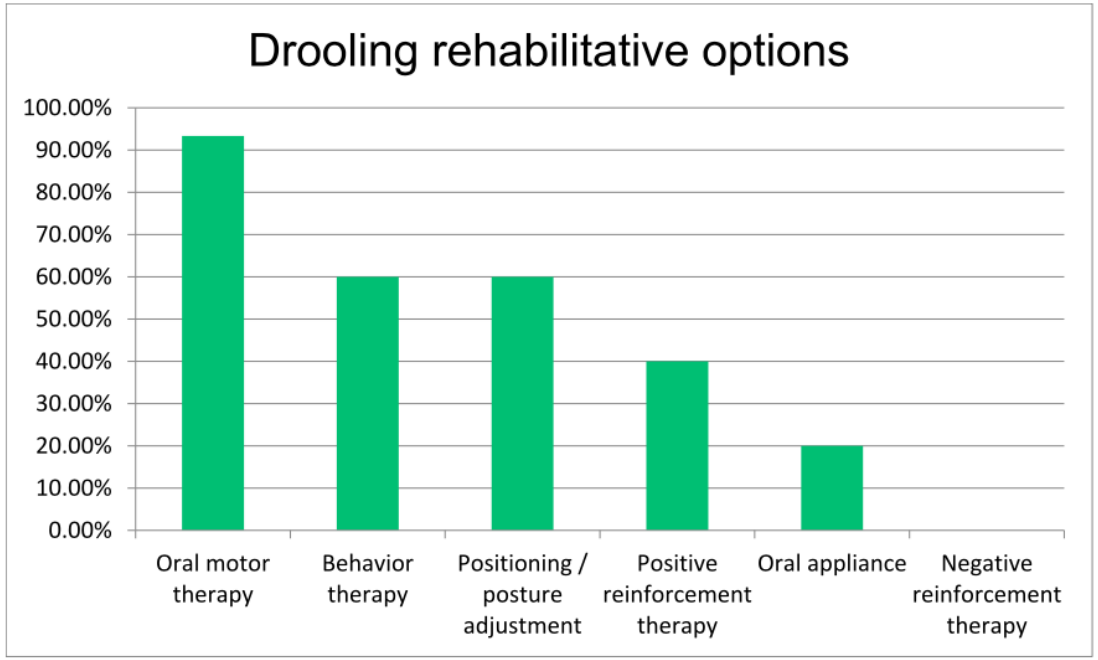
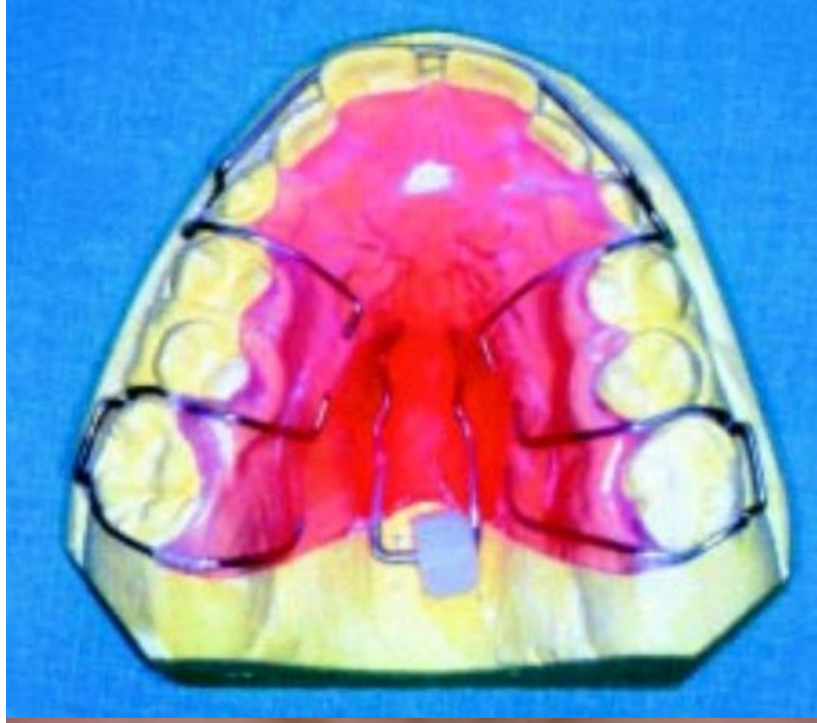


Fig. 1. Rehabilitative options offered for drooling at surveyed IPOG institutions.

Rehabilitative Options

- Intraoral Devices
  - Not recommended in CMC



Pharmacologic Options

- Glycopyrrolate
- Scopolamine patch
- Benzhexol Hydrochloride (Artane)
- Benztropine
- Atropine
- Ipatropium Bromide – not yet studied in children

Medications adverse effects[11]

- Behavioural changes
- Constipation
- Flushing
- Pupillary dilation/visual disturbance
- Urinary retention
- Other less common side effects included
  - Dry mouth
  - Sleep disturbance
  - Vomiting

Glycopyrrolate[12]

Pharmacokinetics

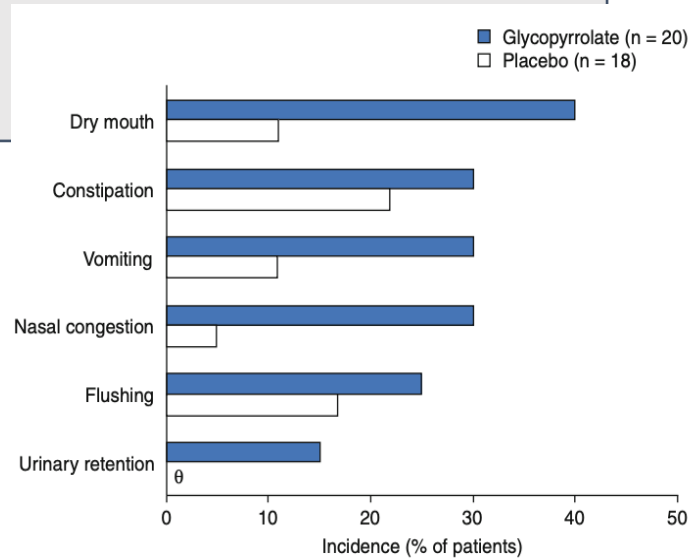
- Time to peak effect 2.5hrs
- Half-life: 3hrs
- Duration of action 8-12hrs

Dosing Regime

- 0.02mg/kg/dose BID (morning and lunchtime)
- Max dose: 0.1mg/kg/dose TID or 3mg/dose TID

Side Effects

- Behaviour changes
- Constipation
- Urinary retention
- Flushing



Scopolamine[11]

Used in nausea/vomiting and sialorrhea

Pharmacokinetics

- Time to peak effect: 24hrs

Dosing Regime

- $^1/_4$ patch q3 days
- Increasing by $^1/_4$ patch every 7 days as tolerated.
- Maximum dose: 1 patch applied every 3 days

Side Effects:

- Skin irritation
- Xerostomia
- Drowsiness

Benzhexol Hydrochloride (Artane)[13]

Used for dystonia and sialorrhea

Pharmacokinetics

- Begins action within 1hr
- Peak action at 1-3hrs
- Duration of action 6-12hrs

Dosing Regime

- 1mg twice daily (morning and lunch/ after school);
- Max dose: 3mg TID

Side Effects

- Behaviour changes
- Constipation
- Urinary retention

Benzotropine[14]

Used for dystonia and sialorrhea

Pharmacokinetics

- Begins action within 1hr
- Peak action at 7hrs
- Duration of action 6-12hrs

Dosing Regime

- 0.02 to 0.05 mg/kg/dose 1 to 2 times daily
- Max dose 4mg/dose

Side Effects

- Anticholinergic effects
- Hyperthermia
- Psychiatric effects (confusion, depression, psychosis)

Atropine[15]

1% solution drops

Pharmacokinetics

- Begins action within 30min
- Peak action at 3hrs
- Duration 5hrs

Dosing Regime

- 0.5mg per drop
- Up to q4hrs
- Max dose 3mg/day

Side Effects

- Anticholinergic effects
- Tachycardia

Medication summary

No consensus in most effective medication(s) nor optimal dosing

Side effect profiles should be considered

Glycopyrrolate, scopolamine/hyoscine, trihexyphenidyl/benzhexol, benztropine, and atropine all effective

Botulinum Neurotoxin[16]

Toxin A and Toxin B

No consensus on:

- Injection strategy
- Dosing
- Time interval between doses

Side Effects

- Dysphagia
- Dysarthria
- Increased salivary viscosity

Botulinum Neurotoxin[16]

- Indications
  (1) Not tolerate/ contraindications to medical management
  (2) Persistent symptoms post medical and rehabilitation management
  (3) Pulmonary complications despite other medical management
  (4) Failed ductal ligation (recanalization)
  (5) If systemic medication or surgery not desired (parental choice or anesthesia contraindication).

- Up to 80% experience reduction in drooling

Assessment for Surgery[17]

Table 2
Important conditions to check prior to drooling surgery.

Associated neurological condition and its future progression
Presence of a muscular disorder
Metabolic disorders
Swallowing status
Pulmonary status
Coagulation status
Presence of sleep apnea
Tonsillar and adenoidal hypertrophy
Macroglossia
Occlusion issues, Dental health, oral hygiene
Number of aspiration pneumonias/respiratory complications
Presence of laryngo-pharyngeal or gastric reflux
Current list of medication
Presence of a gastrostomy tube
Presence of a nasogastric tube
Previous treatment for drooling
Family situation and caregiver
Mental status and ability to consent

Surgical Options[17]

Table 3
Surgical options for drooling performed by IPOG members (an institution can perform more than one type of intervention).

Submandibular glands excision only	68%
Submandibular glands excision + ligation of parotid ducts	50%
Submandibular duct relocation ± sublingual gland excision	40%
Submandibular and/or parotid ductal ligation	33%
Submandibular and/or parotid duct ligation + sublingual gland excision	12%
Tympanic neurectomy	12%

Surgical Options[17]

Table 2. Results Summary

Characteristic	No. of Studies	Subjective Success Rate (95% Confidence Interval), %
Overall	59	81.6 (77.5-85.7)
Mean follow-up duration		
≥1 year	42	83.9 (78.6-89.1)
<1 year	17	76.6 (68.9-84.4)
Surgical procedure		
BSM duct rerouting	21	84.4 (77.7-91.1)
BSMG excision and bilateral parotid duct rerouting	8	87.8 (80.5-95.1)
BSMG duct rerouting and BSLG excision	8	71.5 (63.6-79.4)
BSMG excision and bilateral parotid duct ligation	9	85.2 (78.6-91.7)
4-Duct ligation	4	64.1 (27.6-100)

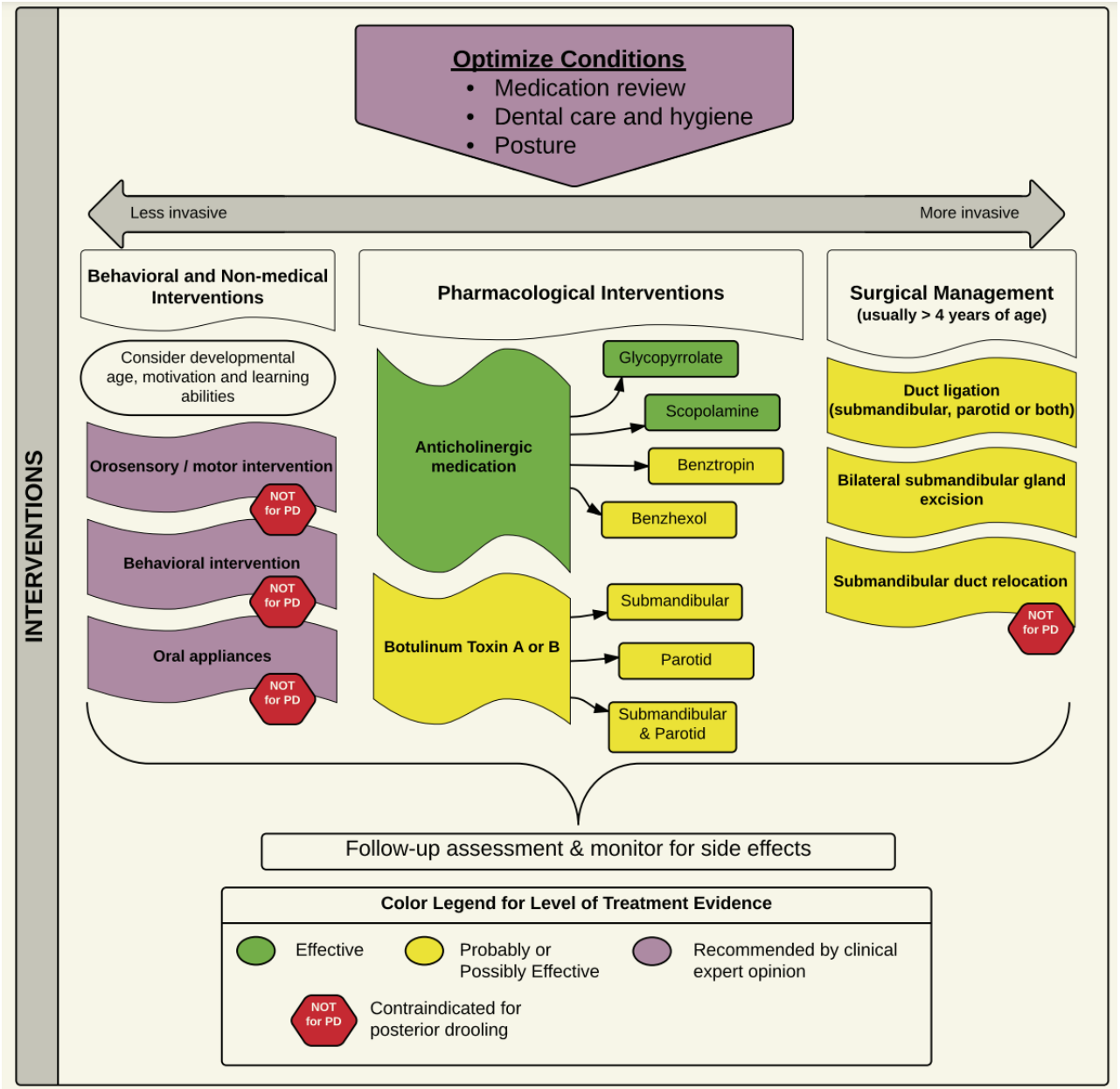
Abbreviations: BSLG, bilateral sublingual gland; BSM, bilateral submandibular; BSMG, bilateral submandibular gland.

Surgical Complications

Table 4
Potential drooling surgery complications.

Submandibular gland excision	Submandibular duct relocation	Duct ligation
Risk of anesthesia	Risk of anesthesia	Risk of anesthesia
Bleeding/hematoma	Bleeding/hematoma	Bleeding/hematoma
Infection	Infection	Infection
Pain (less)	Pain	Pain
Xerostomia	Less risk of Xerostomia	Xerostomia
Dental caries	Less risk of dental caries	Dental caries
Halitosis	Less risk of halitosis	Halitosis
Failure to achieve desired effect	Failure to achieve desired effect	Failure to achieve desired effect
Lingual nerve injury, rarely hypoglossal	Lingual nerve injury	
Marginal mandibular facial nerve injury	Risk of aspiration, to be prevented through careful patient selection	
	Ranula (some remove the sublingual glands to reduce this risk)	Ranula, ductocele, sialocele
Dysphagia secondary to xerostomia		Dysphagia secondary to xerostomia
	Floor of mouth swelling, gland swelling/pain/infection in case of ductal injury or stenosis	Gland swelling, pain, or infection
Hypertrophic scar, keloid	Intraoral wound dehiscence	Intraoral wound dehiscence
		Recanalization leading to relapse

Treatment Options Summary[4]



Case

- Clarify goals
- Re-assess asthma diagnosis and explore alternatives to clobazam
- Treatment indicated
- Consider ~~rehabilitation therapies~~ and medications
- Consider botulinum injections/ surgery in the future
- Counsel on side effects

Take Home Message

- Sialorrhea can have significant physical, mental, and social implications
- Assessment tools exist to formally monitor sialorrhea and treatment response
- Therapies include rehabilitative means, pharmacologic, and surgical options
- Consider side effect profile and alternative indications when selecting medical management

Thank You

Dr. Catherine Diskin

References

1. Hussein I, Kershaw AE, Tahmassebi JF, Fayle SA. The management of drooling in children and patients with mental and physical disabilities: a literature review. Int J Paediatr Dent. 1998 Mar;8(1):3-11. doi: 10.1046/j.1365-263x.1998.00055.x. PMID: 9558540.

2. Fairhurst CB, Cockerill H. Management of drooling in children. Arch Dis Child Educ Pract Ed. 2011 Feb;96(1):25-30. doi: 10.1136/adc.2007.129478. Epub 2010 Jul 30. PMID: 20675519.

3. Hung SA, Liao CL, Lin WP, Hsu JC, Guo YH, Lin YC. Botulinum Toxin Injections for Treatment of Drooling in Children with Cerebral Palsy: A Systematic Review and Meta-Analysis. Children (Basel). 2021 Nov 25;8(12):1089. doi: 10.3390/children8121089. PMID: 34943284; PMCID: PMC8700360.

4. Sialorrhea in Cerebral Palsy. AACPDM Sialorrhea Care Pathway Team: L Glader (team lead), C Delsing, A Hughes, J Parr, L Pennington, D Reddihough, K van Hulst, J van der Burg. Available at: https://www.aacpdm.org/publications/care-pathways/sialorrhea.

5. You P, Strychowsky J, Gandhi K, Chen BA. Anticholinergic treatment for sialorrhea in children: A systematic review. Paediatr Child Health. 2021 Aug 27;27(2):82-87. doi: 10.1093/pch/pxab051. PMID: 35599670; PMCID: PMC9113838.

6. Meningaud JP, Pitak-Arnnop P, Chikhani L, Bertrand JC. Drooling of saliva: A review of the etiology and management options. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;101(1):48–57

7. Sforza E, Onesimo R, Leoni C, Giorgio V, Proli F, Notaro F, Kuczynska EM, Cerchiari A, Selicorni A, Rigante D, Zampino G. Drooling outcome measures in paediatric disability: a systematic review. Eur J Pediatr. 2022 Jul;181(7):2575-2592. doi: 10.1007/s00431-022-04460-5. Epub 2022 Apr 20. PMID: 35441248; PMCID: PMC9192436.

8. Reid SM, Johnson HM, Reddihough DS. The Drooling Impact Scale: a measure of the impact of drooling in children with developmental disabilities. Dev Med Child Neurol. 2010 Feb;52(2):e23-8. doi: 10.1111/j.1469-8749.2009.03519.x. Epub 2009 Oct 15. PMID: 19843155.

9. van Hulst K, Lindeboom R, van der Burg J, Jongerius P. Accurate assessment of drooling severity with the 5-minute drooling quotient in children with developmental disabilities. Dev Med Child Neurol. 2012 Dec;54(12):1121-6. doi: 10.1111/j.1469-8749.2012.04428.x. Epub 2012 Oct 25. PMID: 23094939.

References

10. Morales Chávez MC, Nualart Grollmus ZC, Silvestre-Donat FJ. Clinical prevalence of drooling in infant cerebral palsy. Med Oral Patol Oral Cir Bucal. 2008 Jan 1;13(1):E22-6. PMID: 18167475.

11. You P, Strychowsky J, Gandhi K, Chen BA. Anticholinergic treatment for sialorrhea in children: A systematic review. Paediatr Child Health. 2021 Aug 27;27(2):82-87. doi: 10.1093/pch/pxab051. PMID: 35599670; PMCID: PMC9113838.

12. Garnock-Jones KP. Glycopyrrolate oral solution: for chronic, severe drooling in pediatric patients with neurologic conditions. Paediatr Drugs. 2012 Aug 1;14(4):263-9. doi: 10.2165/11208120-000000000-00000. PMID: 22646067.

13. Jilani TN, Sabir S, Sharma S. Trihexyphenidyl. [Updated 2023 Feb 17]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK519488/

14. Ahuja A, Abdijadid S. Benztropine. [Updated 2022 Nov 7]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK560633/

15. McLendon K, Preuss CV. Atropine. [Updated 2023 Jun 23]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK470551/

16. Ghazavi M, Rezaii S, Ghasemi M, Azin N, Reisi M. Botox injection in treatment of sialorrhea in children with cerebral palsy. Am J Neurodegener Dis. 2023 Jun 15;12(3):97-102. PMID: 37457841; PMCID: PMC10349299.

17. Reed J, Mans CK, Brietzke SE. Surgical management of drooling: a meta-analysis. Arch Otolaryngol Head Neck Surg. 2009 Sep;135(9):924-31. doi: 10.1001/archoto.2009.110. PMID: 19770427.