

Oral Findings in Pediatric Patients with Short Bowel Syndrome

Hasler S^{1, 2}, Chen Z², Choi M^{1, 2}, Alhazmi H⁴, Stamm D^{1, 3}, Culbreath K^{1, 3}, Keefe G^{1, 3}, Nes E^{1, 3}, Duggan C^{1, 3}, Modi B^{1, 3}, Jaksic T^{1, 3}, Sulyanto R^{1, 2}

Boston Children's Hospital, ²Harvard School of Dental Medicine, ³Harvard Medical School, Boston, MA, and ⁴Umm Al Qura University, Makkah, Saudi Arabia



BACKGROUND

Short bowel syndrome (SBS) is a gastrointestinal malabsorptive condition that results from either (1) the loss of function or (2) the resection of a large segment of the small intestine. It is the leading cause of intestinal failure, which is the inability to maintain energy, fluid, electrolyte, or micronutrient balances through a conventional PO diet¹. For this reason, all patients diagnosed with SBS require IV or enteral supplementation to maintain their nutrient and hydration status^{2,3}.

The most common causes of SBS in children are (1) necrotizing enterocolitis and (2) congenital intestinal anomalies, such as mid-gut volvulus, atresia, or gastroschisis⁴.

Clinical manifestations of the SBS population vary widely. Gastrointestinal complications include chronic diarrhea, gastric hypersecretion, dysmotility, anastomotic ulcers, and small intestinal bacterial overgrowth. The most common nutritional deficiencies include fat soluble vitamins (A, D, E, K), iron, vitamin B12, and calcium^{5,6}. Aberrations in vitamin D and calcium homeostasis can lead to metabolic bone disease and rickets⁷. Other sequelae include renal complications, neurocognitive delays, and feeding difficulties secondary to oral aversion, among others⁸. A study exploring the oral health of patients of adults on home parenteral nutrition in the United Kingdom found that patients had a greater instance of dental decay compared to national averages⁹.

Management of patients with SBS includes close monitoring by a team of physicians and nutritionists with frequent laboratory testing. Initial treatment involves parenteral nutrition (PN) to maintain an adequate nutritional status, prevention of fluid and electrolyte abnormalities, and pharmacological management of gastric hypersecretion¹⁰. Long term goals are to maximize enteral feeds with appropriate vitamin supplementation.

Though the systemic management of children with SBS has been studied extensively, there is no existing literature on the dental management of these patients.

OBJECTIVES

This study aims to characterize oral health conditions in pediatric patients with short bowel syndrome (SBS) in order to establish specific guidelines for their oral and dental management.

METHODS

Patients with SBS were recruited from the Boston Children's Hospital Center for Advanced Intestinal Rehabilitation (CAIR). Findings were compared to healthy controls recruited from the BCH Dental Department as well as national data regarding caries prevalence (NHANES, 2015-2016).

Inclusion Criteria

SBS Population

- Age 6 months to 12 years (primary and mixed dentition)
- Severe intestinal failure diagnosed by age 30 days
- At least 90 days' duration of parenteral nutrition which was introduced before 90 days of age Control Population
- Age 6 months to 12 years (primary and mixed dentition)
- ASA I or II
- Low caries risk

Data Collected

- Clinical exam
- Unstimulated salivary pH and buffering capacity
- Questionnaire (birth, medical, SBS, and dental history + home hygiene/dietary habits)

Chi-square tests were used to compare clinical findings between the short bowel and control populations. Regression analyses were completed to determine the relationship between systemic and dental findings.

RESULTS

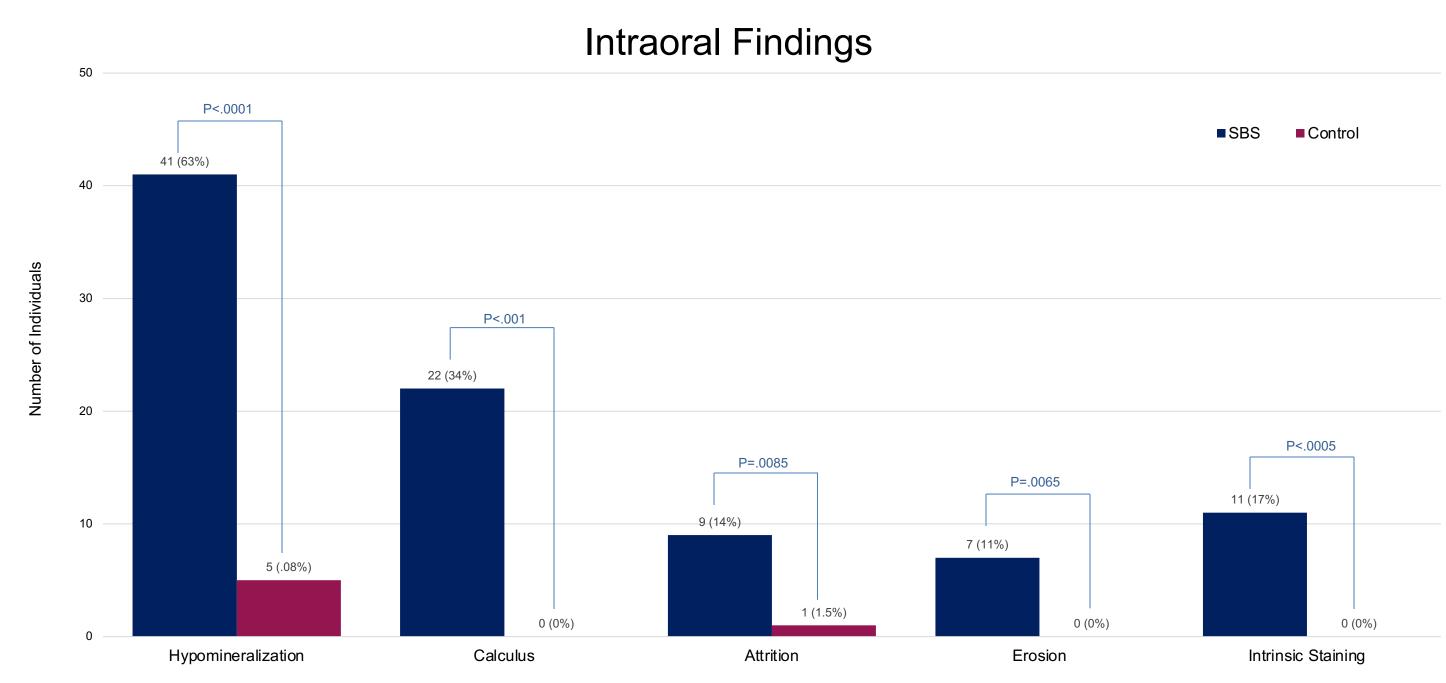


Figure 1. Prevalence of Intraoral Findings. Hypomineralization was the most prevalent oral condition in patients with SBS followed by calculus, intrinsic staining, erosion, and attrition. All conditions were significantly more likely to be found in the SBS population compared to healthy controls: hypomineralization(P<.0001), calculus(P<.001), intrinsic staining(P<.0005), erosion(P=.0065), and attrition(P=.0085).









Figure 2. Clinical Photographs. (a) Hypoplasia of maxillary incisors, generalized intrinsic staining of all primary dentition, and generalized yellow calculus accumulation (b) Generalized yellow calculus accumulation (c) Mild hypomineralization of maxillary posterior dentition and moderate hypomineralization of maxillary incisors (d) Generalized attrition and erosion of maxillary dentition with flattened occlusal surfaces, generalized plaque and calculus accumulation, and plaque induced gingivitis.

RESULTS

65 subjects were enrolled at a median age of 5.9 (3.3-8.0) years.

Intraoral Findings

The following findings were significantly more prevalent in subjects with SBS relative to healthy controls

- Hypomineralization (P<.0001, χ2)
- Calculus Accumulation (P<.001, χ2)
- Erosion (P=.0065, γ2)
- Attrition (P=.0085, χ2)
- Intrinsic Staining (P<.0005, χ2)

Children with SBS did not exhibit increased caries prevalence compared to national data. There was no significant differences in the prevalence of soft tissue pathology between SBS population and healthy controls.

pH and Buffering Capacity

There were no significant differences in pH between SBS and healthy controls (p=.066, SD=.09, t test) or significant differences in buffering capacity between SBS and healthy controls (p=.611, t test).

Relationship Between Systemic and Oral Findings

- Patients who were born extremely pre-term (<28 weeks) were significantly more likely to have moderate hypomineralization (P=.036).
- Patients who are completely or partially g-tube dependent were significantly more likely to have calculus (P=.024).
- Patients with oral aversions were significantly more likely to have calculus (P=.049).

Parental Concerns

- The number one concern parents had regarding their child's oral health was related to the appearance of the dentition (29%).
- 19% of parents were worried about their child's dental and gingival health.
- 9% of parents were concerned for difficulty with home care.
- 6% of parents reported difficulty accessing care with a knowledgeable dental provider.

CONCLUSIONS

- Oral health conditions are highly prevalent in our cohort of SBS patients compared to healthy controls.
- While the prevalence of caries was not increased in this age group, high rates of hypomineralization, erosion, and attrition may increase the risk of developing caries upon the introduction of PO cariogenic foods. Additionally, the lack of available radiographs may underrepresent the true caries prevalence in this population.
- High rate of calculus accumulation may lead to periodontal disease and signals the need for increased periodicity of dental care.
- Both medical and dental providers should offer anticipatory guidance regarding oral health conditions and regular dental evaluations should be encouraged

REFERENCES

- O'Keefe S.I. Ruchman Al. Fishbein T.M. Jeejeebhov K.N. Jennesen P.B. Shaffer J. Short howel syndrome and intestinal failure: consensus definitions and overview. Clin Gastroenterol Henatol. 2006. Jan: 4/1):6-10. doi: 10.1016/j.coh.2005.10.002. PMI
- 16431298.

 2. Pironi L, Arends J, Baxter J, Bozzetti F, Peláez RB, Cuerda C, Forbes A, Gabe S, Gillanders L, Holst M, Jeppesen PB, Joly F, Kelly D, Klek S, Irtun Ø, Olde Damink SW, Panisic M, Rasmussen HH, Staun M, Szczepanek K, Van Gossum A, Wanten G, Schneider
- Yang CF, Duro D, Zurakowski D, Lee M, Jaksic T, Duggan C. High prevalence of multiple micronutrient deficiencies in children with intestinal failure: a longitudinal study. J Pediatr. 2011 Jul;159(1):39-44.e1. doi: 10.1016/j.jpeds.2010.12.049. Epub 2011 Feb PMID: 21324480; PMCID: PMC3112274.
 Squires, Robert H., MD, Duggan, Christopher, MD, Teitelbaum, Daniel H., MD, Wales, Paul W., MD, Balint, Jane, MD, Venick, Robert, MD, Rhee, Susan, MD, Sudan, Debra, MD, Mercer, David, MD, Martinez, J, Andres, MD, Carter, Beth A., MD, Soden, Jas
- Squires, Robert H., MD, Duggan, Christopher, MD, Teitelbaum, Daniel H., MD, Wales, Paul W., MD, Balint, Jane, MD, Venick, Robert, MD, Rhee, Susan, MD, Sudan, Debra, MD, Mercer, David, MD, Martinez, J. Andres, MD, Carter, Beth A., MD, Soden, Jason, MD, Horslen, Simon, MD, Rudolph, Jeffrey A., MD, Kocoshis, Samuel, MD, Superina, Riccardo, MD, Lawlor, Sharon, MBA, Haller, Tamara, BS, Kurs-Lasky, Marcia, MS, & Belle, Steven H., PhD, MScHyg. (2012). Natural History of Pediatric Intestinal Failure: Initial Report from the Pediatric Intestinal Failure Consortium. The Journal of Pediatrics, 161(4), 723–728.e2. https://doi.org/10.1016/j.jpeds.2012.03.062
 Ubesie AC, Kocoshis SA, Mezoff AG, Henderson CJ, Helmrath MA, Cole CR. Multiple micronutrient deficiencies among patients with intestinal failure during and after transition to enteral nutrition. J Pediatr. 2013;163(6):1692-1696.
- doi:10.1016/j.jpeds.2013.07.015
 6. Jeffrey Yang, Chi-fu, AB, Duro, Debora, MD, Zurakowski, David, PhD, Lee, Michele, AB, Jaksic, Tom, MD, PhD, & Duggan, Christopher, MD, MPH. (2011). High Prevalence of Multiple Micronutrient Deficiencies in Children with Intestinal Failure: A Longitudinal Study. The Journal of Pediatrics, 159(1), 39–44.e1. https://doi.org/10.1016/j.jpeds.2010.12.049
- Study. The Journal of Pediatrics, 159(1), 39–44.e1. https://doi.org/10.1016/j.jpeds.2010.12.049

 7. Neelis, Rijnen, N., Sluimer, J., Olieman, J., Rizopoulos, D., Wijnen, R., Rings, E., de Koning, B., & Hulst, J. (2017). Bone health of children with intestinal failure measured by dual energy X-ray absorptiometry and digital X-ray radiogrammetry. Clinical Nutrition (Edinburgh, Scotland), 37(2), 687–694. https://doi.org/10.1016/j.clnu.2017.02.014
- 9. Lee, Gabe, S.., Nightingale, J.., & Burke, M. (2012). Intestinal failure and home parenteral nutrition: Implications for oral health and dental care. Clinical Nutrition (Edinburgh, Scotland), 32(1), 77–82. https://doi.org/10.1016/j.clnu.2012.05.01
 10. Ekema, Milianti, S., & Boroni, G. (2009). Total parenteral nutrition in patients with short bowel syndrome. Minerva Pediatrica, 61(3), 283–291.