

CASE REPORT

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Osteopathic Manipulative Treatment Plus Phototherapy in the Management of Neonatal Hyperbilirubinemia: A Case Report

Abstract

Neonatal hyperbilirubinemia (NH) is costly and has the potential for complications. Standard of care for mild-moderate NH is phototherapy. Osteopathic manipulative treatment (OMT) is safe and effective in other pediatric conditions, but there is little literature on the safety and efficacy of OMT in NH. We present a case of an otherwise healthy 4-day-old term female readmitted for NH and placed on phototherapy. Osteopathic structural exam showed dysfunctions in the following regions: head, cervical, sacral, pelvic, upper extremity, and abdomen, which were treated with osteopathic cranial manipulative medicine, myofascial release, and visceral manipulation. Patient's bilirubin level dropped 3.5mg/dL following OMT, and she was discharged sooner than anticipated than if her NH had been managed with phototherapy alone. According to literature, the median length of stay (LOS) for NH in the United States is 48.00 hours. Her LOS was 24.90 hours. No adverse events were noted. Compared to phototherapy alone, phototherapy plus OMT may lead to improved NH patient outcomes, such as shorter LOS, while still being safe. However, this is a case report of one patient, so no causality can be determined, and more research is needed.

Introduction

Approximately 35,000 infants in the United States (US) are re-hospitalized annually following their birth hospitalizations for treatment of neonatal hyperbilirubinemia (NH), costing about \$361 million per year.¹ NH is elevation in serum bilirubin levels during the neonatal period.² Complications of NH include bilirubin encephalopathy (reversible) and kernicterus (irreversible), as bilirubin is neurotoxic at high levels for prolonged periods of time.²

Although there are both physiologic and non-physiologic causes of NH,² this case report focuses specifically on a patient with physiologic NH. Non-physiologic NH typically involves elevated direct (conjugated) bilirubin levels and usually results from genetic and anatomic abnormalities, such as Rotor syndrome and biliary atresia.² Non-physiologic NH is often more severe than physiologic NH;² however, non-physiologic NH will not be further addressed here, as it is out of the scope of this case report. Physiologic NH typically involves elevated indirect (unconjugated) bilirubin levels and is usually the result of breakdown of fetal hemoglobin in the liver as it is replaced with adult hemoglobin.² Bilirubin, a byproduct of this process, is subsequently

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Disclosures

None reported.

Keywords

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excreted, mostly via stool, with minor amounts via urine.² This process may overwhelm the newborn's liver, resulting in physiologic NH.² Physiologic NH is often less severe than non-physiologic NH, but can lead to the aforementioned complications if not diagnosed and treated promptly.² Risk factors for physiologic NH include feeding difficulties, family history of physiologic NH, and constipation.²

Standard of care for mild-moderate physiologic NH (physiologic NH will be referred to simply as "NH" henceforth) is phototherapy, which uses fluorescent light to facilitate the conversion of bilirubin into forms more readily excretable.² Phototherapy is generally considered safe but has known complications including diarrhea, rash, and disruption to feeding and parent-newborn bonding.² Retinopathy can also occur if proper eye protection is not employed.² According to a recent dataset analysis of 53,259,758 term infants hospitalized for NH in the US between 2002 and 2017, the median LOS for NH was 2 days (48.00 hours).³

While osteopathic manipulative treatment (OMT) is safe and effective for neonates with prematurity⁴ and feeding difficulties⁴ and infants with constipation,⁵ there is little literature regarding OMT and NH. Vismara and colleagues (2019) showed that OMT plus routine care reduced the transition time from nasogastric to oral feeding compared to routine care alone in preterms, $n = 35$ in both groups, total $n = 70$, $p = 0.042$.⁴ Belsky and colleagues (2020) described a case of OMT resolving vincristine-induced constipation in an infantile fibrosarcoma patient.⁵ Cerritelli and colleagues (2014) safely performed OMT on >2,000 neonatal intensive care unit patients.⁶ No adverse events were attributed to OMT in any of these studies.^{4,5,6}

This case report describes a patient with mild-moderate NH managed with a unique osteopathic approach, including OMT to address underlying anatomic and physiologic (structural and functional) aberrations believed to contribute to NH, in addition to conventional phototherapy. Currently, OMT is not routinely included in the management of NH.

Report of Case

Patient was a healthy female born at 37 weeks, 4 days gestation to a healthy 31-year-old G3P2012 mother via uncomplicated vaginal delivery in 08/2021. Mother's entry to prenatal care was delayed until 22 weeks gestation because the family had recently moved to the

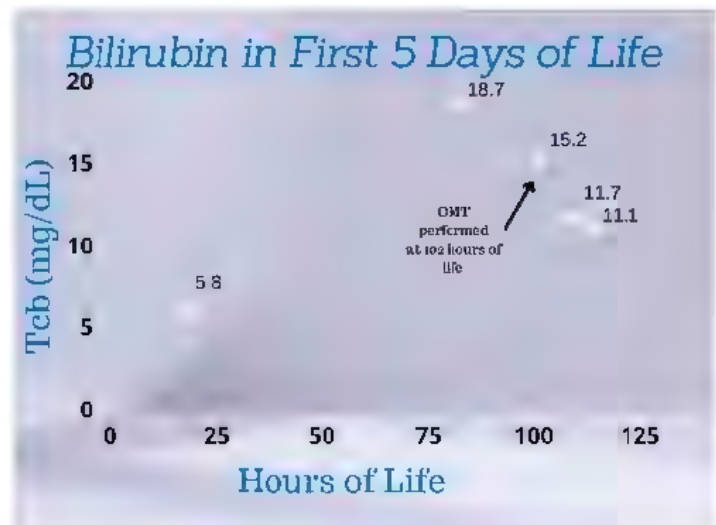
area. Prenatal course was otherwise unremarkable. Prenatal screen was pan-negative. Mother's only medications during the pregnancy were routine prenatal vitamins. Mother denied any tobacco, alcohol, or illicit drug use during the pregnancy. Active labor length was 4 hours, 48 minutes. Anesthesia was epidural. Presentation was direct occiput anterior. Apgar scores were 8 and 9 at 1 and 5 minutes, respectively. Birth weight was 2,925g, which is the 25th percentile. No resuscitation was required other than routine suctioning. She was exclusively breastfed. Poor latch was noted during nursery stay. She also had a family history of a sibling with NH who required phototherapy.

Patient was discharged home with parents on postpartum day #1. Weight was 2,830g, 18th percentile, down 3.25% since birth. She was not jaundiced. Transcutaneous bilirubin (TCB) level prior to discharge was 5.8mg/dL at 19 hours of life (Figure 1), which was low-intermediate risk for her age.

Patient was seen in her pediatrician's office at 82 hours of life and was jaundiced. Parents reported persistent difficulty latching. Weight was 2,679g, 9th percentile, down 8.41% since birth. Total serum bilirubin (TSB) level was 17.5mg/dL, TCB equivalent = 18.7mg/dL, which was high risk for her age. She was readmitted and started on phototherapy under 4 lights for NH. Of note, TSB is the gold standard for monitoring bilirubin levels in NH patients. However, because she was clinically stable, monitoring her bilirubin levels primarily via TCB, which does not require a blood draw, was reasonable.

Parents requested osteopathic neuromusculoskeletal medicine (ONMM) consult during readmission for

Figure 1. Patient's TCB levels in the first 5 days of life. OMT occurred at 102 hours of life. Patient TSB level at 82 hours of life was converted to TCB using the conversion formula found in Reference #7. Original work by Chelsy M. Stephenson, DO.



persistent difficulty latching and constipation. Patient's last bowel movement (BM) was approximately 2.5 days before readmission. The patient had risk factors for NH: feeding difficulties, family history of NH, and constipation.² TCB level prior to OMT was 15.2mg/dL at 102 hours of life, which was intermediate risk for her age. Parents gave consent for OMT evaluation and treatment. Vital signs prior to OMT were within normal limits. Weight was 2,705g, 10th percentile, down 7.52% since birth. This was the only weight taken during her readmission.

Physical Exam

General: Awake and alert, rare crying but easily consolable, no acute distress.

Head: Normocephalic/atraumatic, anterior and posterior fontanelles open and soft, no eye discharge, external ears normal bilaterally, nares patent, mucous membranes moist, poor suck reflex.

Heart: S1 and S2 present, regular rate and rhythm, no murmurs, rubs, or gallops.

Lungs: Clear to auscultation bilaterally.

Abdomen: Soft to palpation and nontender, umbilical stump present.

Genitourinary: Normal female, anus patent.

Musculoskeletal: Normal, equal movement of all four extremities.

Neurologic: Rooting reflex intact bilaterally, Moro reflex intact bilaterally, grip intact bilaterally.

Skin: Warm, dry, jaundiced.

Osteopathic Structural Exam

Head Region: Cranial base compression, bilateral (B/L) occipitomastoid (OM) membranous articular strain, and right condylar compression, treated with osteopathic cranial manipulative medicine (OCMM).

Cervical Region: Occipitoatlantal (OA) joint right facet restriction and C6-C7 restriction, treated with myofascial release (MFR).

Sacral Region: Left S1-S3 compression, treated with MFR.

Pelvic Region: Right intraosseous restriction, treated with MFR.

Upper Extremity Region: Right clavicle restriction and left scapulothoracic restriction, treated with MFR.

Abdomen Region: B/L hemidiaphragm restriction and liver capsular restriction, treated with MFR and visceral manipulation, respectively.

OMT occurred at 102 hours of life.

Patient passed flatus while OMT was in progress. Shortly after OMT, she had a large BM, and her latch improved considerably. Seven hours post-OMT, TCB was rechecked and found to be 11.7mg/dL at 109 hours of life, which was low risk for her age. Another TCB at 113 hours of life was found to be 11.1mg/dL, which was also low risk for her age. She was discharged 24.90 hours after readmission. During readmission, she received phototherapy continuously, except for brief intermittent pauses for breastfeeding. This corresponds to 89 to 114 hours of life. She was not jaundiced at discharge. She did not need to be readmitted again. No adverse events were noted. She only received OMT once.

At patient's follow-up visit with her pediatrician at 8 days of life, she was well-appearing without jaundice. Parents noted that she was feeding well and having regular BMs. Weight was 2,863g, 18th percentile, down 2.12% since birth. At her 4-month well-child check, she was gaining weight appropriately at the 34th percentile, had changed to exclusively formula-feeding, and had no signs of long-term complications of NH. Also of note, there was some speculation that she met failure to thrive (FTT) criteria. While there are multiple well-recognized criteria for neonatal FTT, her weight never measured below the 5th percentile, nor did she cross 2 major percentile lines on the Center for Disease Control (CDC) Girls (0-36 Months) Growth Chart.⁸ Hence, by these metrics, she did not meet FTT criteria. Also of note, the reason for the switch from exclusively breastfeeding to exclusively formula-feeding was her mother's breastmilk production had ceased.

Because this study is a case report, institutional review board approval for it was not necessary.

Discussion

We present a case of a patient readmitted with NH managed with phototherapy plus OMT. Following OMT, her TCB level dropped 3.5mg/dL. She was discharged sooner than anticipated than if her NH had been managed with phototherapy alone and experienced no adverse events. The median LOS for NH in the US according to literature is 48.00 hours.³ Her LOS was 24.90 hours. Rate of TCB decline pre-OMT = 0.18mg/dL per hour. Rate of TCB decline post-OMT =

0.37mg/dL per hour.

To the best of our knowledge, no research addresses the possible mechanism of action (MOA) of OMT with respect to NH. However, existing research suggests that OMT affects the autonomic nervous system, which consists of the sympathetic nervous system (SNS) and parasympathetic nervous system (PNS).⁹

With respect to NH, we hypothesize that OMT acts through the SNS/PNS to address feeding difficulties, impaired bilirubin conjugation, and constipation to lower bilirubin levels. Feeding is primarily under PNS control via the cranial nerves (CN), particularly CN IX, X, and XII.⁹ These CN exit the skull near the cranial base, which is often compressed during delivery.⁹ Patient had right condylar compression, which may have been related to her delivery. The sympathetic innervation of the upper gastrointestinal (GI) tract arises from T5-T9, T6-T9 for the biliary system specifically, and the celiac ganglion.⁹ The sympathetic innervation of the hindgut arises from T12-L2 and the inferior mesenteric ganglion.⁹ The parasympathetic innervation of the hindgut arises from S2-S4.⁹ Somatic dysfunction (SD) in these areas can impair function that can be restored via OMT.⁹

Visceral manipulation is hypothesized to act directly on organs exhibiting visceral dysfunction (VD) to restore optimal function to such organs.⁹ Patient had liver capsular restriction, which was treated with visceral manipulation. Visceral manipulation of fascial strain over the liver could theoretically improve liver function, potentially leading to increased enterohepatic circulation and faster resolution of NH.

Based on our proposed MOA of OMT with respect to NH, regions to be evaluated from an OMT standpoint for NH patients should include the following, with treatment of any SD/VD found:

Head: Particularly cranial base and occipital condyles (patient had cranial base compression and right condylar compression).

Cervical: Particularly OA (she had a restriction at the right OA).

Thoracic: Particularly T5-T9 and T12.

Lumbar: Particularly L1-L2.

Sacral: Particularly S2-S4 (she had left S1-S3 compression).

Abdomen: Particularly upper GI tract, biliary system,

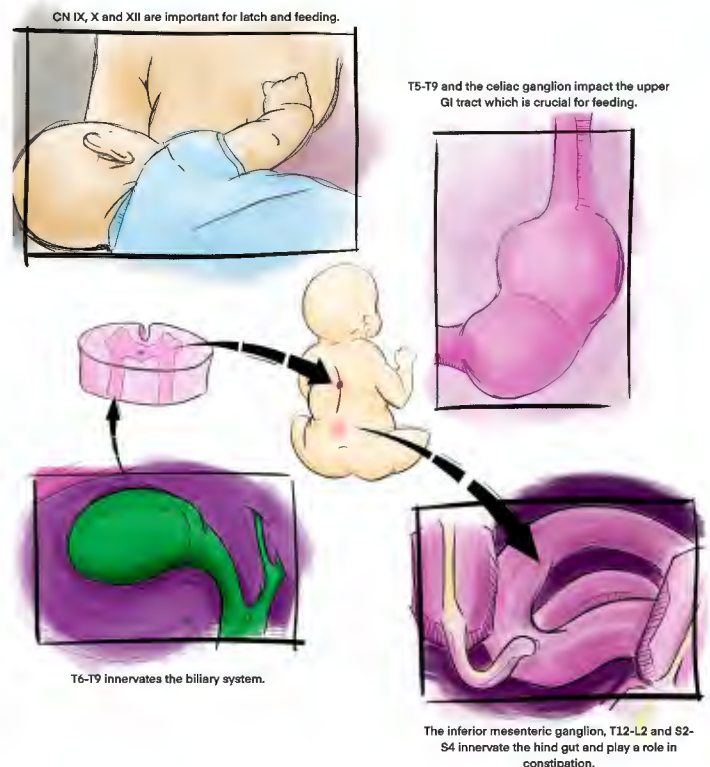
celiac ganglion, and inferior mesenteric ganglion (she had liver capsular restriction) (Figure 2).

Interestingly, she had no thoracic or lumbar SD. However, with respect to NH, special attention should still be paid to these areas during evaluation for the aforementioned reasons, with treatment of any SD found. In summary, this case demonstrates an osteopathic approach utilizing OMT to address underlying anatomic and physiologic aberrations in the form of SD in the ANS and VD in the liver to improve NH, as well as phototherapy.

In addition to patient's shorter-than-expected LOS, another important outcome was the 3.5mg/dL drop in her TCB level following OMT. Her feeding difficulties and constipation also improved following OMT. As previously mentioned, in particular, her latch improved considerably following OMT. Because feeding difficulties and constipation are risk factors for NH,² if they co-present in a NH patient, they should be addressed, as they will impair the resolution of NH if left untreated. Since bilirubin is excreted primarily via stool,² NH patients need to have good oral intake and GI motility to ensure adequate bilirubin clearance. Again, no adverse events were noted.

This study is limited because only one patient is included so no causality can be determined. Also, our case report

Figure 2. Possible autonomic areas in NH. Original artwork by John W. Rajala, DO. The original artwork is based on information found in Reference #9.



did not definitively link this patient's outcome with the proposed MOA of OMT with respect to NH. Additionally, TSB is the gold standard for monitoring bilirubin levels in NH patients, not TCB.

Next steps following our case report could include a study focusing on the most common areas of SD/VD in otherwise healthy term newborns with NH, as needs-based treatment protocols can be considered a limitation from a research standpoint.⁹ This could then evolve into a small pilot study, then a larger cohort study, with the hope of eventually establishing randomized controlled trials. It would also be interesting to delineate the relationship between OMT and bilirubin levels in special patient populations, such as preterms with NH and non-physiologic NH patients. Furthermore, based on the aforementioned anatomic and physiologic premises, OM compression and liver visceral dysfunction, both of which patient had, can be seen as independent risk factors for NH. Seeing how frequently these dysfunctions present in NH patients could also be explored in future studies.

In addition to being potentially efficacious, OMT may be a low-risk addition to phototherapy for otherwise healthy NH patients. Notably, because of this case, our institution has added NH to the list of conditions for which our ONMM service can be consulted.

Conclusion

We present a case of a NH patient managed with phototherapy plus OMT. She had a TCB decline of 3.5mg/dL following OMT, a shorter than expected LOS, and no adverse events. Future prospective studies using TSB should further investigate the relationship between OMT and NH.

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