Torticollis in Infants with a History of Neonatal Abstinence Syndrome

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In this retrospective cohort study, we assessed the incidence of torticollis in infants with a history of neonatal abstinence syndrome. Understanding the elevated risk of torticollis in this population is important for early identification and treatment. *(J Pediatr 2017;■■:■■-■■).*

Neonatal abstinence syndrome (NAS) is a clinical condition that occurs as a result of withdrawal from exposure to opioids in utero. The incidence of NAS has risen almost 5-fold from 2000 through 2012 to a national rate of 5.8 per 1000 live births.[1,2] The opioid epidemic and the widespread use of illicit and prescription opioids in pregnant women has contributed to the increase in NAS. Our region in Ohio has been greatly affected by the opioid epidemic and consequently experienced increased rates of NAS. From 2012 to 2016, 29.4 per 1000 live births had in utero opioid exposure. Among those exposed, 35.1% or 10.3 per 1000 live births were diagnosed with NAS, which is defined in our region as withdrawal symptoms severe enough to require pharmacologic treatment.[3]

There has been considerable effort dedicated to the treatment of opioid-exposed infants after birth and during hospitalization. However, little is known about long-term issues that infants diagnosed with NAS may encounter after they leave the hospital. Some evidence shows that babies with a history of NAS and in utero opioid exposure are at risk for later adverse developmental outcomes including motor and muscle tone problems.[4-6] Torticollis, the postural positioning occurring when the head is twisted and turned to one side, is a condition sometimes seen in infancy.[7] The incidence of torticollis has been reported between 0.3% and 1.9%.[8] Perinatally acquired congenital muscular torticollis is the most common type of torticollis, and it is often thought to be present after birth secondary to intrauterine constraint or birth trauma.[7] Positional or postural torticollis is another subtype of torticollis and has been shown to account for 15%-35% of all torticollis diagnoses.[9,10] This type of torticollis is often associated with plagiocephaly. In our experience following infants with NAS after hospital discharge, we have observed many infants with torticollis and plagiocephaly requiring further intervention and therapy. In our study, we describe the incidence of torticollis in our NAS Follow-Up Clinic population.

Methods

At the Cincinnati Children's Hospital NAS Follow-Up Clinic, we track infants diagnosed with NAS for the first few years of life. Our referral base consists of 14 regional level 2 and 3 birthing hospitals in the Northern Kentucky, Cincinnati, and Dayton areas, which average around 20 000 births per year. All infants who require pharmacologic treatment for NAS at these hospitals are referred to our clinic for ongoing evaluation, developmental follow-up, and continued pharmacologic treatment, if necessary. During clinic visits, infants are assessed by providers and occupational therapists, and if there are areas of concern, referrals for additional therapies are provided.

In our region, we perform universal urine drug testing on all pregnant women at the time of admission to the hospital for delivery. If a woman is positive at the time of delivery or during her pregnancy, urine drug testing and meconium or umbilical cord testing is performed on her infant. Infants are observed and evaluated for withdrawal for a minimum of 72 hours for short-acting opioid exposure and 96 hours for long-acting opioid exposure. All regional hospitals follow standardized treatment protocols for NAS including options for weaning with methadone, buprenorphine, or morphine as first line opioid therapy. When adjunct treatment is required, phenobarbital and clonidine may be used. Infants are treated with opioids exclusively as inpatients, and they are monitored for 48 hours from the last opioid dose prior to discharge. They may be discharged home on adjunct therapy and weaned as outpatients in the NAS Follow-Up Clinic.

In this study, we performed a retrospective review of all infants seen in our clinic with a diagnosis of NAS and a diagnosis of torticollis born between January 2012 and December 2016. This study was approved by the Institutional Review Board of Cincinnati Children's Hospital Medical Center (CCHMC). At each new patient clinic visit, we obtained consent for inclusion in an anonymized research database. Out of 879 patients seen in NAS Follow-Up Clinic during our study period, we were unable to obtain consent in 11 patients, yielding 868 eligible patients. Infants were then excluded if they were <35 weeks gestational age (73 patients), or if they had a major craniofacial abnormality (12 patients). Infants with a diagnosis of torticollis were identified through hospital billing codes.

CCHMC	Cincinnati Children's Hospital Medical Center
NAS	Neonatal abstinence syndrome

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Using a standardized data collection sheet, we abstracted data in the categories of intrauterine exposures, birth measures, neonatal treatments, and postdischarge measures. In utero exposure included short-acting opioids (heroin and prescription opioids), long-acting opioids (buprenorphine or methadone), and polysubstance use (benzodiazepines, cocaine, amphetamines, and marijuana in addition to opioids). Birth measures included sex, gestational age, birthweight, and mode of delivery. Neonatal treatment measures examined opioid and adjunct weaning agents, breastfeeding, length of inpatient stay, and discharge disposition (home with mother, family, or foster care). Postdischarge measures captured at the NAS follow-up encounters included the age at torticollis diagnosis, documentation of associated plagiocephaly, and the requirement of further services for torticollis with early intervention or physical therapy. We conducted a descriptive analysis of the abstracted data reporting counts and percentages for categorical measures and means with SD, medians, and range of values for continuous measures.

Results

A total of 783 infants met inclusion criteria over the 5-year study period. Of those infants with a history of NAS, 87 were diagnosed with torticollis, resulting in an incidence of 11.1% in this sample (see **Figure**).

Characteristics of this cohort of infants are shown in the **Table**. Fifty-eight infants (66.7%) also had a diagnosis of plagiocephaly. Mean age of torticollis diagnosis was 120.9 days (median 103 days). One outlier patient was diagnosed at 582 days with ocular torticollis secondary to his ocular abnormalities. All infants with torticollis were born of singleton pregnancies, and 74.7% were vaginal births. Of these infants, 56.3% were male and 43.7% were female. Mean gestational age was 38.8 weeks with mean birthweight of 2908 g. The majority of these infants were referred for further therapy with 56.3% referred and seen for additional outpatient physical therapy and 28.7% receiving early intervention through Ohio Help Me Grow or Kentucky First Steps.

With respect to in utero exposure, 23% of infants were exposed to only 1 long-acting opioid (12 to buprenorphine and 6 to methadone), with their mothers presumably on and compliant with medication-assisted therapy. Of the infants, 75.9% had exposure to short-acting opioids and 72.4% had exposure polysubstance. Infants were treated with methadone in the majority of the cases (75.9%), and less commonly, buprenorphine (18.4%), or morphine (5.7%). Adjunct therapy was required in 41.4% of infants. Phenobarbital was used in all babies requiring adjunct therapy and in 4 of these cases, clonidine was also used. Length of stay averaged 19.3 days with a SD of 10.3 days.

Discussion

Population-level incidence of torticollis has not been recently reported within the literature, and the most current rate

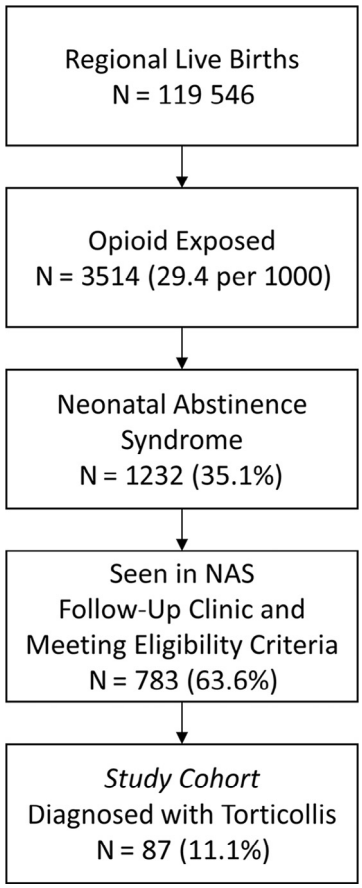

Figure. Flow diagram of infants with NAS seen in follow-up clinic during study time period and diagnosed with torticollis.

of 0.3%-1.9% was published in 1994.[8] Consequently, we identified a comparison cohort within the Cincinnati region to approximate a regional baseline incidence. We identified a patient population who received primary care during their first year of life at 1 of 3 CCHMC outpatient clinics serving the same regional population as our primary cohort during the same study timeframe. Among the patient population of 14 560 infants born from 2012 to 2016 who attended the CCHMC primary care clinics under the age of 1 year, 523 (3.6%) had documented torticollis.[3] This represents a considerably lower incidence than the 11.1% rate identified within the NAS Follow-Up Clinic population.

Since the "Back to Sleep" campaign, there has been an increase in the diagnosis of plagiocephaly seen in infants, and torticollis, typically positional or postural, is the most common associated finding with plagiocephaly.[11,12] With babies sleeping prone on their back, they are more likely to develop flattening of the occiput or plagiocephaly. There is debate about whether torticollis or plagiocephaly is the primary anomaly, but there is consistent correlation between torticollis and plagiocephaly in the literature.[13] This type of torticollis does not appear to involve fibrotic change in the muscle and can be easily treated if caught early.[11] However, if left untreated, true neck contracture may develop along with craniofacial abnormali-

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Table. Characteristics of 87 patients diagnosed with torticollis in the NAS Follow-Up Clinic

Total study cohorts, N	N = 87
Intrauterine exposures	
Any buprenorphine; N (%)	32 (36.8)
Any methadone; N (%)	23 (26.4)
Any short-acting opioids; N (%)	66 (75.9)
Only long-acting opioids; N (%)	20 (23.0)
Polysubstance; N (%)	63 (72.4)
Birth measures	
Patient sex; male, N (%)	49 (56.3)
Gestational age; mean wk (SD), median wk (range)	38.8 (1.7), 39 (35-42)
Birthweight; mean g (SD), median g (range)	2907.7 (460.4), 2845 (1720-3909)
Mode of delivery — Vaginal; N (%)	65 (74.7)
Neonatal treatment	
Weaning opioid — Buprenorphine; N (%)	16 (18.4)
Weaning opioid — Methadone; N (%)	66 (75.9)
Weaning opioid — Morphine; N (%)	5 (5.7)
Adjunct treatment — Clonidine; N (%)	4 (4.6)
Adjunct treatment — Phenobarbital; N (%)	36 (41.4)
Any breastfeeding; N (%)	4 (4.6)
Length of stay d; mean d (SD), median d (range)	19.3 (10.3), 17 (6-73)
Discharged to foster care; N (%)	24 (27.6)
Discharged to kinship; N (%)	21 (24.1)
Discharged to mom; N (%)	40 (46.0)
Discharged to unknown; N (%)	2 (2.3)
Postdischarge measures	
Age at torticollis diagnosis; mean d (SD), median d (range)	120.9 (78.0), 103 (30-582)
Associated plagiocephaly; N (%)	58 (66.7)
Attended physical therapy; N (%)	49 (56.3)
Attended Help Me Grow or First Steps; N (%)	25 (28.7)

ties.[11] Infants with torticollis are at an increased risk for gross motor delay, and those with postural torticollis are at the greatest risk.[10] The nature of torticollis and plagiocephaly in our infants with NAS may be different.

Our study demonstrates an increased incidence in torticollis and plagiocephaly among infants with NAS. In our NAS Follow-Up Clinic population, 11.1% of infants had a diagnosis of torticollis, which is nearly 6 times greater than the general population incidence reported in previous studies and greater than 3 times than is seen in our regional primary care clinics. The majority of these babies also had a co-diagnosis of plagiocephaly (66.7%) with a mean age of diagnosis of 120 days. The subtype of torticollis seen in our NAS babies is likely postural or positional torticollis given the timing of the diagnosis and co-diagnosis of plagiocephaly. Potential explanations for this association could be due to the fact that infants with NAS are often swaddled for calming and they are positioned on their backs or in swings for significant periods of time in the hospital as well as when they are home. The average length of hospital stay for the patients in our study was about 3 weeks. Currently, the babies in our region requiring treatment for NAS are cared for in special care nurseries or neonatal intensive care units and are not rooming in with their mothers and instead potentially spending a significant amount of time lying in hospital beds. Holding and kangaroo care as well as tummy time may be inconsistent in these babies. Infants with NAS also have increased muscle tone in upper extremities as well as neck tightness and lack of head lag. Often the hypertonicity is one of the last symptoms of NAS to resolve and continues to be seen several months after birth. All these factors may lead to infants with NAS developing positional head preferences and cranial asymmetry early in life, which could result in the later diagnosis of torticollis and plagiocephaly.

The healthcare costs associated with treating infants with NAS in the hospital has increased over the last several years.[1,14] Additional healthcare expenditures for infants with a history of NAS after discharge from the hospital has not been estimated but could continue to soar because these children are at risk for ongoing medical and developmental issues. In our study, 28% of infants with torticollis received assistance through state-sponsored early intervention programs and more than one-half required additional outpatient physical therapy for their torticollis, despite exercises and stretching regimens that were provided in clinic. Although we did not look specifically at interventions and treatment for plagiocephaly, referrals to craniofacial specialists and helmet therapy could incur further healthcare costs.

Our study had several limitations. First, we define NAS as requiring pharmacologic treatment for withdrawal from in utero opioid exposure. Infants who were exposed to opioids in utero but did not require pharmacologic treatment are not followed in our clinic and, therefore, we are unable to determine if these infants are also at higher risk for torticollis. Because we were not able to compare our pharmacologically treated NAS babies with those who were opioid exposed but not treated, we are unable to assess if specific factors were more likely to lead to torticollis and plagiocephaly such as in utero exposure, agent of pharmacologic treatment, breastfeeding, or home environment. Although our physician group managed all regional infants requiring pharmacotherapy for NAS during the study period, some infants have been lost to follow-up after initial hospital discharge. Referral for follow-up was not consistent early in our study period. Other potential reasons for infants lost to follow-up include families who moved out of the region and changes in custody of our patients. It is possible that those who did follow-up in our clinic were more likely to have developmental and other problems including torticollis because these babies may have been more difficult to care for at home. However, babies in foster care and in stable home environments are more likely to follow-up in clinic and adhere to recommended anticipatory guidance through stretches and exercises that are provided at every clinic visit, which may be protective against developing torticollis and plagiocephaly. Finally, the type of torticollis was not fully defined in this retrospective cohort.

In conclusion, we found an increased incidence of torticollis and associated plagiocephaly in infants with NAS. Awareness of these potential issues in this population is essential as torticollis may not develop for several months after the infant has been discharged from the hospital. If preventive measures are performed, torticollis and plagiocephaly may be

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avoided. Early detection and treatment might lead to resolution of the problems and might potentially prevent developmental delays. ■

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