NewSTEPs
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Critical Congenital Heart Disease Webinar
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Presenters: Dr. Craig Mason, Careema Yusuf, and Dr. Marci Sontag

Please direct all comments/questions pertaining to this webinar to Thalia Wood at Thalia.wood@aphl.org or 240-485-2701.

Thalia Wood: Now that we've got all that out of the way, recording and muting, I think it is just up 1:00 if we want to get started. Amy, would you like to introduce our speakers for today?

Amy Gaviglio: Yeah, sure. Can you hear me?

Thalia Wood: We can. Thank you.

Amy Gaviglio: Perfect. Thank you all for joining us on this Friday to talk about an issue, I think, that comes up time and time again when discussing point of care and newborn screening conditions, and just how big is data collection and informatics and data integrity. Today, we're going to hear from the original point of care testing side and help … So hearing how that system has addressed issues of data collection and data integrity as well as sustainability of these systems.

We're going to switch over the NewSTEPs who will talk about the data repository and specifically about collection of CCHD cases. We have 3 phenomenal speakers today and I will introduce all of them and then turn it over to the source speaker. We don't want to these webinars to be as discussion-based as possible so please feel free to ask as many questions as you would like.

We really want to make sure that there is an open dialogue and it's not just presentation. As you're listening to the presentation, do think of any questions you would like answered. With that, I will introduce Dr. Craig A. Mason. He is a professor of education and applied quantitative methods at the University of Maine, with interest in informatics, newborn hearing loss and quantitative methods.

Dr. Mason has been involved with the early hearing detection and intervention community for 15 years, working with the US, CDC and various state programs on

multiple projects involving data system design, analysis and long term outcome. He is currently co-chair of the National EHDI Data Committee. My other speakers will be from the NewSTEPs program. First is Marci Sontag who has a PhD and is an associate professor of epidemiology at the Colorado School of Public Health and the associate director of NewSTEPs.

Apparently, that is all she's done but she does a lot more, so that is just a highlight, I guess, of her bio. Then our final and third speaker is Careema Yusuf. She is the manager with NewSTEPs at APHL's newborn screening and genetics department. Her primary responsibilities are related to the Newborn Screening and Technical Assistance and Evaluation Program, or NewSTEPs, data repository, as well as providing technical assistance and support to state newborn screening programs.

Thank you so much to the 3 of you for being here this afternoon. With that, I will turn it over to Dr. Mason and just remind you to hit *7 to unmute.

Craig Mason: Great, thank you. I'm assuming everyone can hear me. Thanks for this opportunity to talk to your group. Probably advance to the next slide. As a background, that over the … Gosh! You think back into the last century now, there was this growing push, I think we saw, for hearing screening, initially, of higher risk infants and then universal newborn screening for all babies.

Then screening for … Expanding and building on that to cover screening for diagnostic and early intervention follow up. This all became … Falls under the umbrella of EHDI or early hearing detection and intervention, and those evolving goals, eventually, were codified in the Joint Committee on Infant Hearing which is known as the 136 recommendations.

If you're unfamiliar with that, what this gets at is the idea that by 1 month of age, all newborn children should receive a screening for hearing loss, preferably before hospital discharge for all hospital birth. Those who do not pass their hearing screen, all infants should then have a formal diagnostic evaluation no later than 3 months of age. Among that subset of those children who have diagnosed hearing loss, they should all be receiving some type of early intervention services no later than 6 months of age.

As you can see from this, EHDI has a little more of an elaborate challenge and elaborate system than some types of screening that can be more … More the work is upfront prior to the hospital discharge. In this case, children often have a second screen that's the outpatient and then you have to work through the process of connecting children and families and collecting data from hospitals to outpatient clinics, to audiologist, to early intervention.

It can be a fairly elaborate and complex process that ultimately requires sharing data and accessing data from different sources at different points in time. Looking at the next slide, what we see is, though, over the last 15 years or so, the EHDI community, nationally, has really done an excellent job at addressing the

challenges that have been faced and improving the number of children going through that process, and successfully completing it.

What I would show here is … Well, what's not shown here is that back in 2000, the number of infants who were being screened was only 52% and latest numbers were up to 98% or more. What this figure shows is that number of infants diagnosed with hearing loss and the corresponding number of those infants that have hearing loss that are actually enrolled in early intervention. We see that since 2000, the number of infants with hearing losses has shown a huge sevenfold increase to over 6,000 children annually based on out latest birth cohort.

Historically, you have to remember that diagnosis, historically, was occurring around 3 years of age or later. You really can't understate the importance that these 6,000 or more kids now that we're seeing are children who, not too long ago, wouldn't have been diagnosed with hearing loss until well after they've missed key developmental milestones.

Similarly, the number of infants with hearing loss that are now enrolled in EI early has shown a nine-fold, well, nearly a nine-fold increase where we're seeing again thousands of children receiving services that are going to help them reach their full developmental potential. If we advance to the next slide, the key to how do we get there, the … And there's been a lot and a lot of it is focused on using and accessing and leveraging data.

A major step in this transition, in the EHDI world was a move from collecting aggregated data to collecting individual child level data, kind of this background. Initially, states were collecting data aggregated from birthing hospitals or birthing facilities. In essence, all the birthing facilities in a state or territory would report on a regular basis. Well, we have … This last month, we have 1,200 births and how many of those births were screened? They report that as well.

You can imagine some of the challenges with that is the data was, at times, lower quality. You'd see situations where hospitals would report, "Well, we have, again, 1,200 births. We screen all our babies," so 1,200 babies were screened. There wasn't a lot of confidence in the accuracy of some of those numbers, so CDC started to push and support and tie to funding a drive to switch from aggregated data to child level data. Looking back at time, there was some question, was that really necessary?

What we saw … Once that transition happened, you did start to see the raw estimates became more accurate, and some of the numbers changed as programs began and hospitals began to realize that there were children being missed. Now, there's more data to actually back that up and identify where that was happening; when it was happening.

Some years ago, to give a sense of the benefits of doing that beyond just that it helps with our raw estimates, it actually helps with trying to look at patterns or

associations that … Well, to give you an example, and I'm a number geek so we'll take a little diversion. You can't ask me to talk about numbers without getting into stats a little bit, in the public health world, you've probably heard of issues like ecological fallacy or Simpson's paradox.

The issue comes down to … What these illustrate are when we're dealing with aggregated data and try to look for patterns or associations, there can be fundamental problems. There was concern that we were seeing this when we were looking at just state aggregated data, even within state. There's a great example that illustrates this, and we're going to take a little diversion to talk about kidney stones and treatment for kidney stones. Let's go to the next slide.

There was a study, years ago, that was looking at these 2 different treatments and the nature of the treatments really aren't important, but they each examined 350 cases of people with kidney stones being treated. When they looked at the aggregated data across these 350 with treatment A, 350 with treatment B, what you saw was that treatment B had a higher success rate, 83% and treatment B reported a successful intervention.

When you start to look at the data more closely, what you saw was that individuals with small stones actually benefited from treatment A. Their success rate was 93% versus 87%. What's really cool or geeky is if you look then at individuals with large stones, they also benefited more from treatment A. In both cases, treatment A actually was better but when you pull the data and only looked at the aggregated data, treatment B looked better.

Now, again, I'm a geek. It's wild when you look at it that way but this can happen in anything when we're dealing with aggregated data. It's not that anybody is doing anything wrong. You can have hospitals that are accurately reporting, "Here's our kids," and when you start looking at subgroups, "Here's our Spanish-speaking families, here's our, our ethnic minority families." When you're only collecting aggregated data and you try to find those sorts of associations, everyone can be doing everything exactly right.

All the data can be reported accurately but it's the nature of the aggregation itself that can lead you, not just to missing associations, but actually drawing the exact opposite conclusion you should be. That's the spooky part of relying on aggregated data, and the need to try to move from … For the space, initially, just the states collecting aggregated data to the states collecting individual child level data, that they avoid making these types of … Again, completely innocent and based on accurate reporting of aggregated data, but still the opposite conclusions you should be.

Let's move to the next slide. As you can imagine, that transition for states, a decade ago, was scary and it required a lot of change. The states needed to come up with new protocols and new agreements. Entirely new data systems have to be developed to deal with the collecting child level data versus just collecting

aggregated counts. The new tools have to come into place. How do you get the hospitals reporting child level data, new mechanisms for collecting the data?

Do you ask nurses to collect it? Can you get it from equipment? We'll talk about that in just a minute. Other backend issues, what about the duplication? Children move from one hospital to another hospital, potentially, prior to their final discharge. How do you avoid counting kids multiple times? To be honest, there were some at the … When we started this process, that felt it was going to be impossible. At least for their state, they may never be able to do that.

Some question whether they might be able to do it for some facilities but not others. It may not be possible to ever get all the data at the child level. Other states felt well, they may be able to get the screening data but getting child level reports from audiologists may be impossible. Certainly, most felt getting early intervention data was going to be impossible, dealing with education systems and other data sources.

There was a lot of concern that would this, in fact, be doable. In fact, I'm moving on, what we saw is that over the next several years, states were able to make that transition. Again, it took a lot of work and everyone should get lots of credit for the energy that was put in, but EHDI has successfully transitioned to now all of the partners are collecting … All of the various programs are collecting child level data starting in 2005.

Or I should say the official reporting for the federal numbers start in 2005. Some states took a little bit longer to be able to report child level data, but everybody is at that point now. One of the benefits that we've started to immediately see from that was having child level data and following individual children from screening to re-screening to diagnosis, to early intervention; allowed you to really follow children through that entire process.

What we were able to see is that where did children get lost at different steps, and what are the factors that may be related to children not completing … Getting lost to follow-up or we say lost to documentation, where they may be getting services, they may be getting diagnosed but there's no official reports of it. For example, if you see a lot … Some states have noted, during this process, that they may see a large loss to follow-up between screening and re-screening.

One solution for that might be scheduling the outpatient screening … Scheduling those re-screenings before the family leaves the hospital the first time. That organizational aspect, maybe play a key role with families getting lost between screening and re-screening. On the other hand, others note loss of follow-up between re-screening, where they have a second outpatient screening and then going on to their formal diagnostic evaluation.

In that case, families might get lost but it's a completely different issue related to access to pediatric audiologists, for example. Maybe there's no pediatric

audiologists that are available within any reasonable distance. Being able to follow individual children over time, you can start to identify where they're getting lost in the process and address specific issues that may be related to their loss to follow-up at each of those different points.

Next slide, and I'm talking longer than I intended, sorry, as you can imagine, accessing the data that's helped to be able to follow children over time has relied on a lot of different technological solutions. Direct equipment transfers is a powerful tool that a lot of states are using, getting the uploads of data on-screen, the results directly from equipment rather than asking nurses to reenter the information somewhere.

Electronic health records is another tool that the EHDI community has been actively involved with trying to access, electronic health records from health systems or directly out-of-hospital health records as a way of, again, leveraging the existence of records. The third party systems have been developed either through commercial vendors or homegrown systems, where hospitals, audiologists; pediatricians can access and enter information directly into an EHDI data system to start to pool all of this child level data.

Ultimately, the issue isn't so much technological as policy. Getting the permissions, getting the agreements in place is eventually, always seems a greater challenge than the technology, so being able to build on whatever existing opportunities or existing agreements that may be in place. For things like CCHD or others that may want to go in this direction, now, maybe they can work off of existing agreements that may be in place, for example, with EHDI as a way of facilitating accessing data.

Next slide, part of what we've seen also with EHDI as we've collected the … As the process has evolved over time, and again, the benefit that we can see with the child level data is that the focus the programs have taken has needed to change over time. If you think, initially, the focus was on screening data and getting all the babies screened. However, only 1 in about 670 kids or new births have hearing loss. I should say again, 1 out of about 670 births have hearing loss based on the latest data.

However, 1 in 7 of the children who fail their screen have hearing loss. Initially, the focus is on screening and that's where we're going to capture most of the babies that have hearing loss. At some point, as the screening rates get really good, there's a transition where more babies with hearing loss … More of the babies out there with hearing loss that aren't being identified are the babies that have … They've failed the screen but now, they're getting lost when they go into diagnosis.

This has been a transition programs have now been working on the last several years; that even though we don't have 100% of the screening, the babies with hearing loss that are being lost are the ones that are failing their screenings but not going on to diagnosis. There's now more of a push and emphasis on that second step in this process, of getting the audiological evaluations, making sure the

families are getting tested, and then getting that data from the service providers.

Next slide, you can actually see this. This is a little piece of a report from Maine where you're able to see some recent data that we have. Back in 2003, about 202 total kids were missed at the screening process but only one of them actually would have been expected to have hearing loss, where there were 40 children that got lost between the screen and diagnostics, diagnostic evaluations.

Out of those 40, 7 of them would have had hearing loss. Because that benefit, understanding the relationship and the math, you can start to see, rather than focus on 202 kids to get 1 child, focus on these 40 kids and you can pick up 7. Next screen, as states work to transition to get the child level data, those same issues that we talked about, the ecological Simpson's paradox and being able to understand things more subtly and follow some of those complications at the state level, it still applies at the national level.

Historically, the data that has been reported nationally, for the national estimates has been aggregated data from the states. To take the next step would be to transition to starting to collect either national, child level data or pool child data level across multiple states. CDC piloted that a few years ago with what's called the iEHDI project where they collected child level data from Indiana, Iowa and Nebraska that gave quarterly child level information, that could then be more accurately pooled across multi [inaudible 00:24:13].

This was done in a way that the children could still be followed over time, but it was all anonymized at the state level. The national dataset didn't contain individual child names but you could follow kids over time. This had a number of benefits. It helped the states with their own data quality, improvement with their data, but also allowed to more accurately look at complex patterns and associations over time. Let's jump to the next slide.

Part of what being able to pool data has done is it's allowed us to … With iEHDI, we're able to look at the differential impact that we saw for different risk factors. We're too small, the numbers are too small to see in an individual state, but as we pooled it across states, we could see. Also looking at loss to follow-up in different urban versus rural settings. We were able to identify how the issues for loss to follow-up in rural communities are different than the issues for families accessing services in urban settings.

Well, let's skip the … I've gone long so we'll skip the developmental outcomes study where 12 states have pooled data to look at some interesting results. I've gone long. Well, we've talked about this. I've already touched on this. Let's keep jumping to the next slide. Finally, I think moving forward, I think leveraging some of the work that's been done in the past, if programs are looking to either collect child level data, start to pool data across states, I think you can benefit from some of … EHDI is set from precedence for sharing data and accessing data, and long term follow-up data.

I think those partnerships can help states to benefit from some of those agreements and that precedence that's been put in place. Also, there may be opportunities to leverage existing data sharing agreements or existing technology, whereas some of the collection tools are already there, either from hospitals or other providers, either partnering with other programs that may be collecting that data or piggybacking systems.

I know several states, for example, are piggybacking their CCHD systems onto EHDI as a way of facilitating and streamlining and then speeding up the process. Why don't we wrap it up there? I've gone long so I apologize to the other speakers. If you have any questions, as you can see, I love talking about this stuff, feel free to contact me directly through … Email is a great way to get hold of me. Again, thank you very much.

Thalia Wood: Thank you very much Craig. That was great. Marci, why don't you go ahead and take over? I'm going to go ahead and see your … Well, you move your own slides. Thank you. You can take over now. Thanks Marci. Marci? Marci, did you unmute your phone? Okay, Marci, we unmuted your phone for you. Hang on everybody. It looks like Marci has to call back in, but [inaudible 00:28:09].

Careema Yusuf: That's okay. We'll go to advance.

Thalia Wood: Okay. We're going to be talking about the NewSTEPs data repository and so Marci is going to talk to you a little bit about what we're collecting, what we're going to try to collect for CCHD, and to just show you what we have right now as far as data points in the repository.

Careema Yusuf: Sure, so maybe we can skip to my section. Good afternoon everyone. This is Careema Yusuf. I was going to talk a little bit about the memorandums of understanding or MOUs that we have here at NewSTEPs. Really, these MOUs are to provide the following information. It talks about data ownership, data sharing … Data reports we'll be providing, and then how we are storing and making the data secure and private and confidential.

Within the NewSTEPs data repository, we will be collecting data on individual case level data for the different confirmed conditions that are found through newborn screening. We wanted to share with you that we are requesting each newborn screening program to sign an MOU with us in order to provide that kind of information. I wanted to just briefly talk about the process for getting an MOU signed. We're very happy to say we have over half of the programs who have signed an MOU.

For those of you who are not sure if your program has an MOU with us, please feel free to contact me. You will see my contact information on the next slide, but going back to the process, it's really simple. We will send you an MOU and in that template, we ask you to make sure you review it and ask us lots and lots of

questions. A lot of programs have different processes of how things get signed within their programs.

Some folks don't like it to be called an MOU but want it to be a data sharing agreement, and we're happy to accommodate that. Then we ask that you make any edits to the template using the Track Changes feature in Word and then send that back to us. I think some of you may have questions around providing CCHD data to us which is a little bit different from the dried bloodspot data that the programs are collecting.

Please feel free again to reach out to me to find out if your state newborn screen program has an MOU with us. We're happy to accommodate if you need an extra signature on your end in order to provide us this point of care information. Here is my contact information. Again, please feel to reach out to me with any questions and [inaudible 00:31:00] with you, either the MOU template or if your program has signed one, the one that they did sign with us. Thank you. Is Marci connected now? She's on. She's trying to call in. There she is. Okay.

Thalia Wood: Marci, are you on?

Marci Sontag: Can you hear me Thalia?

Thalia Wood: Now we can hear you. Thank you Marci.

Marci Sontag: Okay, so sorry about that. I thought I was … Anyway, I was on my computer and it wasn't letting me speak. Thank you all for your patience and I'd like to thank Dr. Mason for presenting the importance of the EHDI data and data collection at both the local and a national level and what we can learn from it. Dr. Mason and I had the privilege of speaking at the CDC Grand Rounds this week and really, it became very apparent, the differences between EHDI and CCHD data collection.

We're all really in the baby stages or infancy stages of data collection for CCHD and I'd like to present to you today on how … One step in moving forward with CCHD data collection. There have been talks about a CCHD registry at a national level and collecting data on, not only the short term outcomes of newborn screening, but then long term outcomes of what happens to these children following identification through screening, newborn screening or one of the other screening mechanisms for CCHD.

There's talk about potential research opportunities, looking at outcomes of these kids and really validating the importance of screening. One of the biggest questions we get at NewSTEPs is how many kids are diagnosed by newborn screening from CCHD every year. That's just something we can't answer yet, and so I want to talk about how we could potentially partner together to answer that, and introduce a NewSTEPs data repository.

Hopefully, many of you have seen our NewSTEPs data repository. It is really geared

towards state and territorial, public health newborn screening program. We have several different modules within that repository we call data and quality indicators, and outcomes of newborn screening programs. 6 of the those quality indicators are … 6 of the 8 quality indicators are directly geared towards the point of care screening outcomes as well such as time to diagnosis and intervention, loss to follow-up, false negatives, etc.

We provide confidential data reports to state newborn screening programs, so this is not meant to be punitive. This is meant to be supportive so that states can understand their own data. We use our data repository to create reports, really, for quality improvement and program management within the state while providing them how their state compares to other state programs. Then we have data on the positive newborn screening, cases who end up with a diagnosis and we have case level data entry, and then metrics related to that.

That's what I'm going to be talking about today. I'm going to start first with what does this look like in other cases. These are from dried bloodspot, disorders that are identified through the dried bloodspot. This is time to confirm the diagnosis, and so we can know how long does it take to diagnose children with each of these diseases following a positive newborn screen. I will tell you now. I'm not going to go into details of these.

This data is about a year old. I'm just showing them to demonstrate the power of what we can get out of this data and say, "Oh, look." For MCADD, we're diagnosing them really pretty early, where for [Gal 00:35:08], that's a little bit later in life and we can then use that for quality improvement activities and so forth. Then I can get into a specific disorder, and I have all the states presented with cystic fibrosis. This is time to release of out of range results, and this is a great way for quality improvement.

Each of these states is blinded but we can look at state 744 since we can get all of their cases diagnosed in a tight timeframe, whereas, say, 736, there's a very wide range there. what we can learn from state 774 to really help those other states? This is the type of information we would like to be able to write for CCHD. How many cases are being diagnosed? What's the timing of their diagnosis? What's the timing of intervention?

Here's time to intervention. Similarly, this is, again, just for cystic fibrosis comparing across many different states, and you can see that time to intervention is very wide; that some states are entering their time to intervention for individual babies. This is at the individual baby level and then aggregated. You can see for 742, it's in the first 2 weeks of life and for state 774 and 810, that intervention might not be happening until the first couple of months of life.

Then finally, here's time to confirm diagnosis. There's lots of details related to what does that mean, to have an intervention in the diagnosis. We are really excited to be able to compare these data and to be able to help states use that for quality

improvement. We can look at the time in this for dried bloodspot screening and you can say, “Well, time from birth to collection is a very important metric for us now in the timeliness of newborn screening.”

You can see this data presented here, birth to collection on the first row, in 2012, was about 35 hours and that has now decreased to 26 hours as a median time from birth to collection of that first dried bloodspot. Could do something very similar for CCHD screening and determine those types of metrics as to when is that screening actually happening, and how is it happening in the state. I’m going to go on to our demo site and I’m going to share my screen to allow you to see our repository. Thalia, can you see the repository?

Thalia Wood: Yes, we can. Thank you.

Marci Sontag: This is a demo site, so this is not … You will not see real data as I go into this site. Hopefully, most of you have access to the NewSTEPs data repository. If not, you can go to newsteps.org and register for the data repository and talk to your state level administrator to give you permission for your state access as appropriate. If I’m logging in here, this front screen is very similar to what you would see in your data repository and I’m going to go up here towards these cases, and enter cases for a given case.

Again, you would only see cases for your individual state and the cases that are entered here are all fictitious. These are just our practice cases. What I’m going to do is I’m going to add a new case to show what this looks like for critical congenital heart disease. Aside from Colorado, I will enter the state of Colorado and I can start over here by typing the condition here. It pops up with CCHD as I just type CCHD or I can also go over to the right side under disorders and type CCHD here.

I continue. We do not collect any identifiers on our end that would allow us to link back to any other data source. This state unique ID is the one that states enter in order to track back to their own when they want to come back and enter more data on this child. Our data depository has undergone review here at the University of Colorado Multiple Institution Review Board as well as we have sought advice through our IRB of OHRP, the Office of Human Research Protection, on this data resource.

Both of the actually said it was okay for us to collect dates of birth, but most of the states we work with have said, “We just can’t supply date of birth.” The data that we’re presenting here, it gives templates to type in the date of birth but nothing is actually saved. It calculates all of the differences in ages for each of these outcomes but doesn’t save the data. You’ll see that as I go through.

A calendar pops up, I’m going to say this baby was born on the 1st of September. I’ll calculate the birth year here in the bold, and this baby was born at 3:08am. I’m going to say there’s [inaudible 00:40:38] in weeks. Birth weight in grams can be entered or not entered. It’s not a required field but you can enter that there.

Biological sex is a female, this was an African American baby, not Hispanic.

Then this here, which newborn screening indicator, this [inaudible 00:41:01] risk for the disorder. This is a artifact of this being part of the dried bloodspot screening, and so we are having internal discussions about how we might be able to change this. The initial screen, "Was prenatal testing done that indicated this infant was at risk for the disorder?" No, but this … And this could be an mechanism to also collect data on infants who were diagnosed following a prenatal screen.

Was there a family history? No, and was this individual diagnosed later in life? This is a false negative. We have that mechanism to collect not only prenatal testing but also, was this a false negative, to be able to track that at the local and national level. The point of care testing information, here, we want to know when was that first screen done. We're not going to collect information on each of the 3 screens, so baby was on the 1st, the screen was done on the 2nd and it was done at 6:20am.

[inaudible 00:42:01] time elapsed from birth is calculated there, is 24 hours. When did that intervention happen? Now, you might ask what is that intervention, and we are actually pulling together a group of cardiologists to make sure we have the right definition of what intervention we want to track here. What we've done for other disorders is the first time someone has made a decision on behalf of that child, it changes the medical care of the child.

That could be in the case of a child with CCHD, that they were started on prostaglandin or they were started on oxygen, something that helps that child related to a CCHD diagnosis. We are working on refining those definitions for you. We say, "Well, you know what? It was actually 2 days of life that the screen … They failed the screen, they had it repeated a couple times. By the time we go that interpreted, because it was on a weekend, it took a couple days."

"We started the baby on oxygen on day 2 of life and we confirmed the diagnosis on Monday." Is this infant receiving … Our final question here, is that infant receiving treatment or care out of state, yes or no? That's just to help state be able to track those border babies that might be going across borders from where they were born. Then our final question for now is what was that diagnostic workup and what was the final diagnosis?

In the case of this child, this was a hypoplastic left heart syndrome. You can enter more than 1 diagnosis here, and that some infants have more than 1 defect. In addition to the work that we're doing for the case definitions, we will be building out additional information that states can enter into this to be able to track what information do we know that this child has a hypoplastic left heart. What were the diagnostic indicators and more information can be entered, that that workgroup is just beginning their work.

Then I'm going to say yes, this data is complete. We know everything we can about that baby and then I'm going to save it. You'll see that the state unique ID is 1235.

I'm going to come back to that here in just a minute. We have all the dates here. When I save, all those dates are not going to be saved. When I go back into this, 1235, and I look at this child again … Let me go back into it as a … In edit mode.

You can see that all of those things in bold are there but none of the confidential information related to birth date are entered. It's pretty straightforward. We're just really now trying to introduce this to the CCHD community, so you all can enter data into the repository. As Careema said, it's guided under an MOU. I'm going to go back to my slide. I have some additional comments here on implementation of the case definitions and I've talked about that a little bit.

Since we only have 15 minutes left in our time together, I would like open it up for questions. I can go into more about these case definitions and how we're doing this and what we're doing with the case definitions. I'd love to have each of you … If you have questions about how to enter the data, the value you see in this. This is step 1. This is not the long term follow up registry, but this will give us that opportunity to be able to enter data and know how many babies were screened, diagnosed in each state, and really be able to measure the impact of newborn screening.

Thalia Wood: Thank you Marci. Just remember to do *7 to unmute your phones and ask a question or you can type a question into the chat box.

Amy Gaviglio: This is Amy. Can you hear me?

Thalia Wood: We can.

Amy Gaviglio: Perfect. My question is, I think, a little bit more for Dr. Mason but maybe also to touch on data definition, first of all, for CCHD. I'm wondering if you can, for this group, clarify the difference between loss to follow-up and loss to documentation as it pertains to EHDI. I do think it's an important distinction, and do you see this is something we should be thinking about for CCHD screening as well?

Craig Mason: Hi, this is Craig. Am I unmuted?

Thalia Wood: You are.

Amy Gaviglio: You're good.

Craig Mason: I think, within EHDI, it's often difficult to differentiate between the 2. The idea for loss to follow-up, it's where the family is completely lost in the sense that you have no idea what's happened to the child. It becomes loss to documentation. It, on one hand, reflects the subset of those cases where children are, in fact, getting services but we don't have documentation of it. Or that the family may not be … You may just not be getting information back on a family.

Generally, the 2 of them … Usually, what happens in practice is that we just don't know what's … The families just fall through the cracks and disappear.

Marci Sontag: Then to add to that, Amy, for CCHD screening, I think it absolutely could be something similar. The difference with CCHD is that these babies are followed … Typically, follow-ups begin before they leave the hospital. Hopefully, they should not be lost to follow-up but it could be that public health never hears about that baby, that they know the baby, through their mechanisms, failed the screen, that they got into care but never found out what that diagnosis was or that diagnosis was confirmed. That's then, I think, loss to documentation and not loss to follow-up.

Amy Gaviglio: I was just thinking maybe a good term for us to think about, because I don't think … Historically, within bloodspot and very similarly, it's not so much that they maybe aren't lost to follow-up. In some cases, yes, but a lot of times, it is that loss to documentation piece, so it may be something we want to think about at term including, especially, in CCHD because I think you're absolutely right.

Hopefully, they're never lost to follow-up and it's purely a, "We can't close the loop from a documentation standpoint." I do have one other question if no one else does. This has to do with … And I think it will talk a little bit about the EHDI piece, but also about the idea of not only collecting data but also entering data or reporting data. Certainly, the CDC has a very robust report for EHDI that they ask for each year.

I'm wondering, Dr. Mason, if you're aware of whether the EHDI world has ever done a cost analysis for programs and how much it costs to perform this level of follow-up. Obviously, with EHDI, I would expect that you're following up on many more kids than we would expect to for CCHD, but I think even that cost analysis and then looking at the state's refer rate could be beneficial to CCHD programs that are looking to build an individual level data collection. I'm wondering if anything like that exists.

Craig Mason: There have been some cost analyses done at different points in time. I would think that [inaudible 00:50:13] at CDC had done one several years ago. I can't remember the exact numbers. It did ultimately prove cost effective, even with hearing screen which you can imagine, there are additional costs given the numbers and the complexity, but especially once you look at any … And that's the value of looking at the long term outcomes.

For EHDI, the reduction … You can have reductions for kids entering special education requiring additional services, etc. For CCHD, I think there's the obvious long term health impacts in that, but if someone's interested, get back to me and I can get you a copy of the latest cost analysis write-ups. I just can't think of exact numbers off the top of my head.

Amy Gaviglio: Perfect, thank you.

Marci Sontag: Craig, you made a comment on the char box about the CCHD data score reporting systems is very cool. Just to remind everyone, this is also available for all of the dried bloodspot screening but it's actually available for EHDI as well. We have built that module. We have not actually done anything with it because EHDI is so well managed by CDC and HRSA program already.

If an individual state wanted to use this for their own tracking, this would be … For EHDI, they'd be able to use that as well.

Craig Mason: I think that the value that you can see by studying the pool of data across states, particularly for smaller states, to benefit from sharing their data with others and being able to understand those long terms impacts, and just associations for different subgroups in that. It's extremely valuable to be able to start doing that. For CCHD, at such a relatively young program, to be already at this level is exciting to me.

Marci Sontag: Well, any time I can hear that from somebody who's such a data guy, that's great. We do have a couple of poll questions. Thalia, if you want to take control back, we're still willing to … Happy to answer questions. I'd love to hear what people think, if this is something that people would be willing to do. This first question is what do you hope to gain from national data for CCHD?

This is a better understanding of effectiveness, what programs are picking up, as probably what the diagnosis are being diagnosed, and then comparison of different protocols. You can go ahead and click on your computer screen.

Thalia Wood: Thank you, and this one is choose your best answer. It's not choose more than one answer, and I'm going to go ahead and see what we're getting as the results here. Well, I'd say there's a clear winner for what people are looking for.

Marci Sontag: It's hard to be able to just pick one though.

Thalia Wood: That's true. The next few questions, you can actually pick more than one. Well, go ahead [inaudible 00:53:49] see the better understanding of effectiveness is the clear winner there. This one is, "What barriers exist that could be addressed at a national level, Reimbursement, specific poll thoughts training, looking at earlier algorithms or connections to public health to health?" Okay, this has been very interesting.

Marci Sontag: Amy, I think you developed this question. Do you mind expanding a little bit or helping us interpret this, the results on this one?

Amy Gaviglio: I know. I was just thinking, "Did I write this question?" I think what I was thinking was, in terms of reimbursement, maybe was more along the lines of how to pay for something like this or resource allocation. Certainly, we know there still does not exist a PCP code for [inaudible 00:55:04] screening in a well-baby without an

associated hypoxemia. If people were thinking about that reimbursement from that perspective, I think I completely agree that that's a big one.

I think, looking at earlier algorithms, I know in our last talk, we heard from the out-of-hospital birth population who've had quite a bit of success with actually screening earlier, so that may help for them. Connections to public health, I think that one's pretty self-explanatory in terms of barriers to robust data collection and who's doing it, and how they've done it, and maybe how we can we can work better together to do that. I guess I'm not too surprised that reimbursement, connections to public health are the highest.

Thalia Wood: Okay. We have 1 more poll question. How do you envision using national data to change your program, the ability to have more data, change the screening algorithms, change follow-up processes or nothing? I'm glad to see nothing is losing.

Marci Sontag: I was like, "Oh gosh! What if everybody says nothing? There's no reason to have national data."

Careema Yusuf: You're being validated Marci.

Marci Sontag: Oh, phew. It's very interesting that changing following processes is running along with changing screening algorithms.

Careema Yusuf: I think this makes sense to me from when I think about how we've used contributing to national data and EHDI. In Minnesota, we certainly have changed the way we follow up from when we first started. Our individual data collection in 2007 till today is completely different in where we put our focus, also in terms of what we've learned about health equity in the follow-up processes. That's cool to see and I think that there is a lot of value to collecting this national data for that purpose.

You do learn a lot about where the gaps are in the system and where to maybe put your time. Similar to what Dr. Mason was saying in terms of you have 200 who've missed screening but only one of those may have a hearing loss, versus a smaller number who missed their diagnosis but more of them are like … They have a hearing loss, so where to maybe put some of your time and effort, especially since many of us are resource-limited is very helpful.

Marci Sontag: I agree and I think this could be really helpful for resource allocation and also for being able to learn from others. If we can learn what's going well in Maine that we can learn to apply in Colorado, that's a win for everybody so we don't need to recreate the wheel. For all of you who are on, I hope that you will … If you're not the person who has the authority to enter data in your state, talk to the people who are. We really would love to know how this works for you.

This is a work under development and if there's questions related to the repository,

Careem, Thalia and myself are all happy to talk with you about that. Please reach out to us. We would love to start collecting those data so we can say, "Look at what an impact all the work we're doing in CCHD newborn screening is having." We do recognize that there are 15 state programs who have no requirement and are not collecting data at the state level, and that's another area that many of us are interested in thinking about.

Are there ways that we can change those requirements and look at those abilities to collect data at the local level within each state, because the only way we'll be able to improve is to actually be able to have the data and measure the metrics.

Amy Gaviglio: That's [crosstalk 00:59:17].

Thalia Wood: Thank you so much.

Marci Sontag: Go ahead Thalia. You can-

Thalia Wood: I was going to say thank you so much, and I know we're right at the top of the hour. I wanted to thank all of you for being on the call. Thanks to our speakers and remind you that this are now quarterly. The next call will be December 9th. Anyone have the last word?

Amy Gaviglio: No. Well, I guess I will have the last word because I'm speaking now, but thank you all, especially to the speakers. I do think there's a lot for us to learn from EHDI and it was, I think, comforting to know that EHDI has gone through this process, and that they've come up the other side with a very robust system. I certainly envision that for CCHD as well. I know NewSTEPs is taking the lead on getting that done so yeah. Thank you all again and wishing everyone a wonderful weekend ahead.

Marci Sontag: Thanks everyone. Thanks Amy.

Thalia Wood: Thank you.

Amy Gaviglio: Thank you.