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Forensic medical evaluations of child maltreatment: A proposed research agenda☆

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ABSTRACT

Physicians play an important role in the forensic evaluation of suspected child abuse and neglect. There has been considerable progress in the medical field, helping distinguish findings related to maltreatment from other conditions or circumstances. Nevertheless, important questions remain. This article covers several of these questions and proposes a research agenda concerning five main topics: sexual abuse, neglect, fractures, abusive head trauma, and physicians work in interdisciplinary settings. The suggestions are hardly inclusive, but offer suggestions the authors think are priorities, and ones that research could reasonably address. By providing some background to gaps in our knowledge, this paper should be of interest to a broader audience than just medical professionals.

Physicians play an important role in the forensic evaluation of suspected child abuse and neglect. There has been considerable progress in the medical field, helping distinguish findings related to maltreatment from other conditions or circumstances. Nevertheless, important questions remain. This article covers several of these questions and proposes a research agenda concerning five main topics: sexual abuse, neglect, fractures, abusive head trauma, and physicians work in interdisciplinary settings.

The article is based on a background paper solicited by the United States Institute of Medicine for their recent report on research regarding child maltreatment. The scope of the paper was limited to a research agenda on medical aspects of evaluating children for possible abuse or neglect. For this reason, many important topics such as prevention are not included. It is clear that there would not be a perfect consensus on what issues should be prioritized. We fully recognize that the topics for the proposed research agenda are hardly inclusive. There is a long list of potentially important topics. Hopefully, our experience as "senior" child abuse pediatricians helped develop a useful agenda. In addition to sharing ideas among ourselves, we requested and incorporated input from approximately twenty other physicians in this field.

Child Sexual Abuse

Most victims of child and adolescent sexual abuse (or assault) present for medical evaluation after they have disclosed sexual contact. Typically, acute and healed ano-genital trauma, sexually transmitted infections (STIs), and forensic evidence are absent. Few children present with ano-genital trauma or an STI as their sentinel injury/condition, and in the absence of a

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sexual abuse history, specificity of the exam finding or the STI is critical to diagnostic accuracy. The diagnosis of sexual abuse or assault relies on the medical history, visualization of ano-genital injury, presence of certain STIs, and other evidence from the appropriate collection of forensic materials. Diagnostic accuracy depends on the differentiation of abusive injuries from non-abusive injuries and medical conditions that mimic trauma. Accuracy also depends on the appropriate interpretation of STIs identified in various sites by various testing modalities. Several research questions are important to improve this process:

Research Related to the Diagnosis or Confirmation of Sexual Abuse

What Does the Medical History Obtained from the Child Contribute to the Clinical Assessment, Medical Diagnosis, Treatment, and Investigation? Diagnostic accuracy in medicine relies upon the history from the patient; nearly 80% of medical diagnoses are made primarily based on patient history (Peterson, Holbrook, Von Hales, Smith, & Staker, 1992). However, there is considerable variability in the extent to which medical providers obtain a medical history from a suspected victim of child abuse: some rely entirely on information gathered during the forensic interview, some rely partially on such information, and some do not rely on this information at all; rather, they obtain an independent history from the child. In child maltreatment, the goals of the medical history are to evaluate the likelihood of abuse and assess symptoms and conditions that affect a child's physical and mental health in order to guide treatment and restore health. When the medical history is gathered for the purposes of medical diagnosis and treatment, that information may be admissible in a legal proceeding pursuant to an exception to the hearsay rule; however, the medical history is not consistently considered as part of the investigation. The diagnostic utility of the forensic interview has been explored (Katz & Hershkowitz, 2012; Lyon, Ahern, & Scurich, 2012). Research addressing the utility of the medical history within the forensic context, including impact on legal outcomes, is lacking. While one study did describe urogenital symptoms reported by children during the medical history that may be specifically associated with sexual abuse trauma (DeLago, Deblinger, Schroeder, & Finkel, 2008), the forensic significance of other physical, emotional and behavioral symptoms has not been described. Due to variability in how health care providers gather and interpret information from children suspected of having been abused or neglected, research assessing the utility of the medical history is problematic. However, studies that compare information gathered from the forensic interview to information gathered during a medical history with respect to impact on clinical assessment, medical diagnosis, treatment, multidisciplinary investigation, and presentation of evidence during court testimony would help address this question.

What is the Diagnostic Validity of Acute and Non-Acute Anal Findings for Abusive Trauma? Anal findings of acute and non-acute trauma that can be attributed to sexual abuse or contact are considered rare and not well described. Few studies have described anal injuries observed in small samples of children (Heppenstall-Heger et al., 2003; McCann & Voris, 1993) and adolescents (Adams, Girardin, & Faugno, 2001) evaluated for sexual abuse. The interpretation of some findings documented in these studies has changed over time. The lack of a true "gold" standard has made it difficult to draw valid inferences about the diagnostic significance of anal trauma in a child or adolescent with a history of anal penetration. However, large scale studies that characterize anal findings and confirm sexual contact by other means, such as recovery of forensic materials or detection of an STI will improve diagnostic accuracy and assessment of anal injuries.

Which Methodologies are Optimal for Detecting STIs at Different Sites (Oral, Genital, Anal, Urine, Blood) in Children and Adolescents? Addressing this question will improve detection of STIs in children and adolescents evaluated for possible sexual abuse/assault. Improved detection will, in turn, improve clinical treatment and provide forensically important information. Some studies (Gallion, Dupree, Scott, & Arnold, 2009; Girardet et al., 2009; Kellogg, Baillargeon, Lukefahr, Lawless, & Menard, 2004) have compared various methodologies including culture, nucleic acid amplification tests (NAATs), wet mount and serology for detecting STIs in children evaluated for sexual abuse. Criteria for testing, however, are not uniform and comparative studies of newer NAATs have not been conducted in this patient population. Large-scale (or multi-center) comparative studies of prepubertal and pubertal children utilizing different testing modalities at multiple sites are needed to further evaluate this question.

What is the Diagnostic Validity of Using Nucleic Acid Amplification Tests (NAATs) to Detect STIs, Specifically Neisseria gonorrhoeae, Chlamydia trachomatis, Herpes Simplex Virus (HSV) Types 1 and 2, and Trichomonas vaginalis? Although data exists to validate detection of some STIs with some NAATs, there are knowledge gaps regarding which NAATs are most specific and sensitive for which sample site (e.g., urine or vaginal NAATs) and type of lesion (e.g., HSV NAATs for vesicles and/or ulcers). In addition, the appropriate use of NAATs may vary with patient sex and age (prepubertal vs. pubertal). As an example, transcription mediated assays (TMA; one type of NAAT) that detect gonorrhea and chlamydia at genital and anal sites have higher sensitivity and specificity than other NAATs, but have been studied primarily in adult populations (Golden, Hughes, & Cles, 2004; Moncada, Donegan, & Schachter, 2008; Schachter, Chow, Howard, Bolan, & Moncada, 2006). Results of studies involving adult populations with low prevalence rates of STIs support use of NAATs in children, but further studies in prepubertal children are needed. Knowledge in this area will improve detection of STIs in children and adolescents evaluated for sexual abuse.

What is the Yield of Testing for STIs in Children of Varying Ages, with Varying Clinical Presentations Suspicious for Sexual Abuse? Rationale have been proposed for targeted (select patient populations based on age, symptoms and type(s) of sexual contact) and sequential (high sensitivity test followed by a high specificity test) STI testing in children and adolescents evaluated for sexual abuse (Ingram, Miller, Schoenbach, Everett, & Ingram, 2001; Palusci & Reeves, 2003; Siegel, Schubert, Myers, & Shapiro, 1995), However, new tests for STIs have changed detection rates; for example, some protocols have recommended testing prepubertal children for gonorrhea or chlamydia only if they have symptoms of infection (Shapiro &

Makoroff, 2006), but more recent studies (Gavril, Kellogg, & Nair, 2012) report that these infections have been diagnosed in asymptomatic prepubertal children. Establishing the clinical indications for testing will improve detection of STIs in children and adolescents evaluated for possible sexual abuse/assault.

What is the Optimal Time Interval within Which Various Types of Forensic Evidence (Linens, Clothing, Orifice Samples, Skin Surface Samples, Hair, Debris) can be Recovered? Clarifying the indications for collecting specific forensic materials based on patient age, gender and clinical circumstances will improve diagnostic and forensic yield. Studies to date are limited and results are varied; one study indicates that collection of body swabs in prepubertal children who present for examinations more than 24 h after sexual contact has extremely low yield (Christian et al., 2000). More recent studies (Young, Jones, Worthington, Simpson, & Casey, 2006; Girardet et al., 2011), however, have indicated that evidence can be recovered beyond 24 h in children. Collection of orifice swabs and blood can be traumatic, particularly for young children, so clarification is needed to establish the optimal clinical approach. Although there is much speculation, no studies have addressed the detection of epithelial cell DNA and interpretation with regards to sexual abuse.

Research on Diagnostic Accuracy Related to Physical Findings

How Does Training and Experience Influence the Assessment and Interpretation of Ano-Genital Findings? Understanding the role of the medical professional's training, supervision and expertise in the detection and interpretation of ano-genital findings is critical to ensuring appropriate child protection, investigation and prosecution. Concerns include whether Sexual Assault Nurse Examiners (SANEs) inappropriately attribute examination findings to trauma, and whether physicians miss trauma or fail to attribute examination findings to trauma (Bechtel, Ryan, & Gallagher, 2008). Additionally the role of experience in diagnostic accuracy has been described (Adams et al., 2012; Gavril et al., 2012), but the number of practitioners in these studies has been small. Differences of opinion do occur between and within groups of practitioners from different disciplines. However, different opinions based on the same sound research are problematic, compromising not only diagnostic accuracy, but patient care and the investigation. The goals of peer review are to educate, build consensus and improve diagnostic accuracy. The format for peer review varies from periodic informal discussions to weekly formal presentations and critiques of written and photo-documentation with article citations. Some peer reviews involve practitioners from one discipline, while others include clinicians of multiple disciplines, training and experience. The impact of peer review (or other approaches) on diagnostic assessment and accuracy has also not been studied or reported.

What are the Anatomical/Developmental Changes within the Hymen/Vestibule from Age 9 Years through Adolescence? Anatomical structures and their normal developmental variations are sometimes confused with trauma, Only one study has described longitudinal changes in hymenal anatomy from birth through age 9 years (Berenson & Grady, 2002). Describing normal anatomical development will improve differentiation of normal variants from changes due to trauma. For example, the interpretations of the narrow hymenal rim, deep hymenal notches, and lateral deep notches or clefts of the hymen have varied over the past several years; clarification of these findings as developmental variations or residua of trauma would improve diagnostic accuracy.

When are Ano-Genital Warts Sexually Transmitted? Current studies describe detection of HPV DNA in newborns, infants and children (Rintala, Grénman, Järvenkylä, Syrjänen, & Syrjänen, 2005), but the relationship between the presence of HPV DNA and development of clinical disease remains unknown. The likelihood of sexual transmission in a child with clinical HPV on the genitals, peri-anal area or oral mucosa among young children is difficult to assess. Similarly, the role of patient age and/or clinical characteristics such as lesion location, subtype(s), and morphology in evaluating the likelihood of sexual transmission is unknown. Addressing these questions will improve the clinician's ability to assess the likelihood of sexual contact in children with HPV disease. In contrast to the subsequent question which focuses on vertical transmission of STIs, this question focuses on the differentiation of vertical (transmission during or preceding birth), horizontal-non-sexual (hand-to-anogenital contact during hygiene or bathing care) and sexual modes of transmission that result in clinical HPV.

What is the Prevalence and Persistence of STIs Acquired Perinatally? Maternal STIs may be transmitted to the child's oropharynx, genitals or anus before or during birth. Some infections produce immediate symptoms in the newborn, while others, such as HPV and Chlamydia may produce delayed or no clinical symptoms. Some studies (Bell et al., 1992; Sinclair, Woods, Kirse, & Sinal, 2005) have reported prolonged latency periods for clinical manifestations of STIs acquired during birth, but presence of both maternal disease and transmission to the newborn was not confirmed. Assessing the likelihood of sexual transmission of an STI diagnosed in an infant or child depends on knowledge of perinatal transmission, including which sites are typically infected, and the latency periods for each STI.

Research over the past 10 years has significantly impacted the interpretation of ano-genital findings, forensic evidence collection protocols, and testing for STIs. Medical providers are ideally suited to talk with children about their abusive experiences, but the information gathered during this encounter is not consistently utilized for investigation by law enforcement or child protective services professionals. Opportunities for further understanding and improvement remain, particularly regarding STI detection, testing modalities, and protocols for testing.

Child Neglect

Neglect is the most prevalent form of child maltreatment, involving over two-thirds of reports substantiated by child protective services (CPS) (U.S. Department of Health and Human Services, 2014). Despite sounding benign, neglect can have

severe and often long-term sequelae (Dong, Dube, Felitti, Giles, & Anda, 2003; Dong et al., 2004; Dube et al., 2001; Dubowitz, Papas, Black, & Starr, 2002; Eckenrode & Kendall-Tackett, 1996; Teicher et al., 2004). At the same time, physicians and other professionals often grapple with the question of what constitutes child neglect. Addressing the following definitional and/or diagnostic issues should substantially enhance practice and children's health, development and safety.

What is the Likelihood and Nature of Harm Resulting from Circumstances that are Thought to Jeopardize Children's Health and Wellbeing?

Most state laws include the risk of harm in their definitions of neglect (DePanfilis, 2007). This element is especially important to understand, because many neglectful situations do not result in immediate harm. Nevertheless, there is the potential for longer term harm. Addressing potential harm may serve a valuable role in helping prevent ongoing neglect and possible adverse outcomes. Estimating the likelihood and severity of future harm is often difficult, There are areas where epidemiological data help, such as the increased risk of a serious head injury from a fall off a bicycle when not wearing a helmet (Wesson, Spence, Hu, & Parkin, 2000). Assessment of possible neglect would be enhanced by knowing the likelihood of harm and the nature of outcomes for situations such as failure to thrive, obesity, lack of immunizations, poor hygiene, being home alone, unsafe sleep practices in infants, alternative healthcare/cultural approaches such as acupuncture for asthma, and drug exposure in newborns. Additionally, clarifying the harm attributable to certain conditions or circumstances would be useful (e.g., what is the nature of harm resulting from prenatal and/or postnatal cocaine or marijuana exposure?). In sum, such data would help guide clinical assessments that too often are loosely based on subjective views. That said, there are circumstances that reasonably can be considered neglect, without requiring such evidence. Examples include a 4-year-old home alone or a child who is chronically hungry.

What is Known about the Preventability of Circumstances Thought to Constitute Neglect?

Neglect is generally understood to be the result of an omission in parental care, care that if provided would have prevented actual or potential harm. The extent, however, to which specific circumstances are attributable to a lack or inadequacy of care and are therefore preventable is often uncertain. Examples include serious injuries to children not in motor vehicle restraints (i.e., to what extent would a suitable restraint have prevented the injury?), pulmonary conditions or complications in children exposed to second and third hand smoke, diabetic ketoacidosis and poor dietary control, accidental drowning of young children, and Sudden Infant Death Syndrome (SIDS) in children sleeping in the prone position. It is evident that the degree of preventability falls on a continuum (Webster & Starnes, 2000). On one end, the drowning of an infant in a bathtub or a toddler in a swimming pool is completely preventable. Alternatively, sleeping in the prone position reduces but does not eliminate the risk of SIDS. One study assessed the preventability of deaths in children denied regular medical care because of parental religious beliefs (Asser & Swan, 1998). Knowledge of the degree of preventability will enable appropriate determinations of neglect and guide the development and implementation of preventive measures at both individual and community levels.

How do Physicians Decide to Invoke the "Neglect" Label?

Physicians' decisions regarding possible child maltreatment can have far reaching consequences, including referral for investigation, parental drug testing and placement of children outside the home in addition to the provision of in-home services, therapeutic day care and parenting classes. Practice concerning neglect appears to be especially variable, in part because of a lack of definitional consensus. Unlike physical and sexual abuse that involve acts of commission, the acts of omission in neglect are inherently challenging to assess and adjudicate. Physician biases that have been referred to elsewhere in this paper may be even more influential in determining neglect. It is unclear how physicians weigh various factors in their assessment of possible neglect, including possible bias concerning race and socioeconomic status, their views of what constitutes neglect, characteristics of neglect such as the type and chronicity, differences in state laws, their reluctance to "judge" and antagonize families, the potential for harm as well as other factors. Understanding physician decision-making regarding neglect should help develop a more uniform and optimal approach to practice. This could be achieved, for example, by a shared understanding of the potential for harm.

What is the Yield of Medical Evaluations for Children Suspected of having been Neglected?

As mentioned earlier, neglect is the most prevalent problem for children in the child welfare system. Some children are reported to CPS because their medical needs have been neglected (i.e., medical neglect). Others are reported for other forms of neglect, and they have significant medical problems (e.g., severe asthma) that may be related to neglect. It seems reasonable that both these groups be medically evaluated as part of a neglect investigation, but it appears that few are. It would be useful to know the yield of such evaluations. This question also pertains to the remaining majority of children reported for neglect. One form of neglect is often accompanied by other forms of neglect (Dubowitz, Pitts, & Black, 2004). For example, a child who is poorly supervised may also not have received adequate healthcare. Another consideration is that many of the conditions involved in neglect, such as poor household sanitation and little food in home, may affect health. In

addition, neglect seldom involves just one child in a family or household; there is reason for concern that others too have been affected (Hines, Kantor, & Holt, 2006). Thus, information on the yield of medical evaluations for these vulnerable children would guide policy and practice by helping develop criteria for which children and circumstances should be prioritized for a medical evaluation.

What Approaches are Effective for Evaluating Different Forms of Possible Neglect?

Research could help clarify effective approaches to evaluate different forms of neglect. Inadequate food and impaired growth offer a helpful example. Medical professionals routinely track children's growth and diagnose when it is faltering (i.e., failure to thrive), perhaps due to neglect. But children, particularly older ones, may experience hunger regularly and yet have normal or accelerated growth. A brief questionnaire that probes food insecurity and possible hunger is especially useful (Kleinman et al., 2007). Another example concerns the assessment of the adequacy of supervision, a common concern (Anderst, Dowd, Schnitzer, & Tryon, 2012). Further research is needed to find effective ways to assess possible neglect, parent–child interaction, and medical neglect associated with poorly controlled diseases (e.g., asthma, diabetes).

Fractures

Fractures are common pediatric injuries, and skeletal trauma in physically abused children represents a small percentage of all childhood fractures. Of those fractures that result from abuse, most are found in infants (Day, Clegg, McPhillips, & Mok, 2006; Kemp et al., 2008; Leventhal, Martin, & Asnes, 2008). Abused infants and children may present with skeletal trauma as their sentinel injury, and fractures are regularly identified during the medical evaluation of suspected abuse as well as other conditions. The timely identification of skeletal injury can lead to earlier identification of abuse, sparing the victim further-sometimes life-threatening-injury (Ravichandiran et al., 2010). Alternately, there are a number of pediatric diseases that cause skeletal fragility and fractures early in life that can be misdiagnosed as child abuse (Bishop, Sprigg, & Dalton, 2007). To improve early identification of abusive injuries and appropriate consideration of medical conditions that may mimic abuse, priority should be given to answer the following research questions:

Research to Improve Early Identification of Skeletal Injury

What is the yield of Skeletal Survey in Children of Various Age Groups and with Different Presenting Signs or Symptoms? Radiographic skeletal survey is the standard screening tool for detecting clinically unsuspected fractures in possible victims of child abuse. Presently, the American Academy of Pediatrics recommends radiographic skeletal survey for children younger than 2 years of age with fractures and other injuries suspicious for child abuse (Flaherty, Perez-Rossello, Levine, & Hennrikus, 2014). These recommendations are subject to interpretation by physicians tasked with identifying abused children, depending on what a clinician thinks is suspicious. Many abused children present to medical care with non-specific symptoms that are not easily identified as being the result of abuse, leading to missed opportunities for diagnosis (King, Kiesel, & Simon, 2006). A recent analysis of more than 700 consecutive skeletal surveys performed at one children's hospital revealed occult skeletal trauma in more than 10% of those tested, influencing the diagnosis of abuse in over half of the positive cases (Duffy, Squires, Fromkin, & Berger, 2011). Infants younger than 6 months, those who presented with ALTEs, seizures, or other signs of abusive head trauma had the highest rates of positive skeletal surveys. Not all children with seizures or ALTEs were imaged, however, and the criteria for obtaining skeletal surveys in those who were imaged are unknown. Further research to identify clinical presentations that warrant skeletal survey will provide clinicians with more specific recommendations for screening and improve the early detection of child abuse.

What is the Yield of Skeletal Survey in Children of Various Age Groups Who Present to Care with a Single Fracture? Because current guidelines for obtaining skeletal surveys are non-specific, and the evidence on which recommendations are based is limited, significant variation exists in how skeletal surveys are obtained in clinical practice. Race and socioeconomic status appear to influence physician practice in obtaining skeletal surveys when children present with skeletal trauma, leading to both over and under-reporting of abuse in different populations (Lane & Dubowitz, 2007; Lane, Rubin, Monteith, & Christian, 2002). For example, minority toddlers with accidental fracture are 5 times more likely to have a skeletal survey ordered than their Caucasian counterparts (Lane et al., 2002). Similar differences by race and socioeconomic status are reported for infants who present with skull fractures (Wood, Christian, Adams, & Rubin, 2009). Additionally, the risk of child abuse likely varies by both age and the specific bone that is injured. Prospective studies with large samples would be valuable to identify clinical scenarios that require skeletal surveys and those that do not. Additional data may improve screening for abuse, safety for those abused, and refine the indications for screening and lessen unnecessary testing.

What is the Yield of Follow up Skeletal Surveys in Different Clinic Presentations? Repeating skeletal surveys 2–3 weeks after an initial presentation of suspected abuse improves diagnostic sensitivity and specificity for identifying skeletal trauma in abused infants (Harper, Eddleman, & Lindberg, 2013; Kleinman et al., 1996; Zimmerman, Makoroff, Care, Thomas, & Shapiro, 2005). Not all abusive fractures (e.g., rib and metaphyseal fractures) are visible radiographically initially, and prospective studies have shown that repeat skeletal imaging increases the number of fractures diagnosed by more than 25% in abuse victims (Kleinman et al., 1996). More recent data suggest that repeating the skeletal survey will identify skeletal trauma in 10% of infants whose surveys were initially negative (Bennett, Chua, Care, Kachelmeyer, & Mahabee-Gittens, 2011). Furthermore,

repeat skeletal surveys can assist in dating of injuries, clarify questionable findings, and can alter the clinical diagnosis in equivocal cases (Harper et al., 2013; Kleinman et al., 1996). Research has also shown that for children with equivocal findings on an initial skeletal survey, repeated radiographs can both confirm and exclude fractures, and change the ultimate diagnosis (Zimmerman et al., 2005). The benefits of repeating skeletal surveys have been established, but further research is needed to identify the appropriate indications for doing so. These may include children with multiple fractures, those with other injuries concerning for abuse, children with equivocal findings on skeletal survey, and for those with certain non-skeletal injuries. Presently, there are no established guidelines for repeating skeletal surveys, and large, prospective studies that include broad populations of children with suspected abuse will help establish data to inform needed standards.

What are the Most Sensitive, Specific and Cost-effective Methods for Identifying Skeletal Injuries in Abused Children? In addition to the standard radiologic skeletal survey, a number of alternative and supplemental imaging modalities, such as MRI, PET scans and ultrasound, have been used to identify skeletal trauma in abused children. In choosing the optimal radiologic strategy, considerations including the sensitivity and availability of the study, radiation dose, cost (including operator time, financial and sedation costs), the availability of qualified radiologists for interpreting pediatric images, and sedation requirements. New radiologic modalities have been used with varying success in the early identification of skeletal trauma in abused infants (Drubach et al., 2010; Kemp et al., 2006; Perez-Rossello, Connolly, Newton, Zou, & Kleinman, 2010; Wootton-Gorges et al., 2008). Establishing the safest, most reliable modalities will improve diagnostic testing and the early recognition of abuse.

Research Related to the Differential Diagnosis of Skeletal Injuries

What are the Indications and Yield of Testing for Metabolic Bone Disease for Children Who Present with Unexplained Fractures? Which Tests are Needed in Different Clinical Scenarios? There are diseases and conditions that affect collagen and/or bone mineralization that can mimic skeletal trauma due to abuse; identifying these diseases or conditions is critical to reducing false accusations of abuse (Bishop et al., 2007). Vitamin and mineral deficiencies, and genetic and infectious diseases can all mimic abuse (Byers, Krakow, Nunes, & Pepin, 2006; Marquardt, Done, Sandrock, Berdon, & Feldman, 2012; Shore & Chesney, 2013; Taylor, Chaudhuri, Davis, Novelli, & Jaswon, 2008). Although recommendations have been published recently by the American Academy of Pediatrics (Flaherty et al., 2014) clinical practice is quite varied. Because some of these diseases are uncommon, large studies are needed to determine the characteristics that best identify children with underlying metabolic bone disease, the yield of screening tests for metabolic bone disease in children with different fracture patterns and age groups. This knowledge will help reduce false accusations of abuse, limit unnecessary testing, and will improve diagnostic certainty in identifying abused children.

When does Vitamin D Insufficiency Become Clinically Significant in Causing Fractures? Vitamin D deficiency is a common problem in infants and young children. There is controversy regarding the role of vitamin D insufficiency and deficiency in fracture susceptibility in infants (Botash, Sills, & Welch, 2012). A few researchers have offered suboptimal vitamin D status as an alternative explanation to abuse in infants and young children with unexplained fractures (Keller & Barnes, 2008; Paterson, 2009). Other researchers have examined the relationship between suboptimal vitamin D levels and fracture susceptibility, finding little support that suboptimal Vitamin D explains fractures in abused children. Chapman et al. (2010) reviewed 40 infants and young children with rickets for fractures, and found that all the identified fractures were in those children with radiographically abnormal bones, and none sustained fracture types that are more specific for abuse. Schilling, Wood, Levine, Langdon, and Christian (2011) examined vitamin D levels in infants and young children presenting with fractures, and found no association between vitamin D insufficiency and children with multiple fractures or those who were abused. However, neither of these studies compared fracture rates of infants with suboptimal vitamin D with those who are vitamin D sufficient. Furthermore, because vitamin D deficiency and rickets represents a continuum of laboratory, clinical, radiographic and pathologic abnormalities, further research is needed to determine where in the spectrum of deficiency does fracture susceptibility become clinically significant (Marquardt et al., 2012). If Vitamin D insufficiency in the absence of clear laboratory or radiographic support of metabolic bone disease does increase fracture susceptibility, pathological fractures may be misdiagnosed as abuse. Conversely, abused children may be put at further risk if inflicted fractures are inappropriately attributed to Vitamin D insufficiency. Further research of the relationship between vitamin D and fractures will improve diagnostic certainty in identifying child abuse, and reduce false accusations of abuse, and provide opportunity to treat a preventable form of bone disease.

What are the Clinical Indications for Screening for Osteogenesis Imperfecta (OI), and Which Methodologies are Best Used for Testing? OI describes a group of genetic disorders defined by skeletal fragility and recurrent fractures. Like other metabolic bone diseases, some children with OI are thought to be victims of abuse prior to being diagnosed with metabolic bone disease (Byers et al., 2006). Early studies attempting to differentiate OI from abuse suggested that clinical examination by an experienced clinician was as sensitive as biochemical testing in differentiating the two (Steiner, Pepin, & Byers, 1996), but later research found that clinical examination alone misses a few children with OI (Marlowe, Pepin, & Byers, 2002). It has become evident that there is great genetic complexity in the molecular basis of OI, and single genetic abnormalities can display significant phenotypic variation (Cundy, 2012). In the past decade, the genetic bases of 10 new OI variants have been discovered, and for some disorders, the clinical spectrum of disease has not been completely described (Marlowe et al., 2002). Given these advances, further research is needed to provide clinical guidance for when to test, appropriate testing methodologies (biochemical, genetic, both), and interpretation of genetic mutations. Such information will help protect

abused children and reduce false accusations of abuse. Additional research is needed to clarify the clinical presentations of mild–moderate and new forms of OI, and thus improve diagnostic accuracy.

What Role Does Prematurity Play in Fracture Susceptibility? Physiologic and metabolic processes arising from prematurity predispose premature infants to bone fractures. It has been estimated that up to 24% of very low birth weight infants sustain fractures, although reported frequencies are quite variable (Carroll, Doria, & Paul, 2007). Fractures in premature infants may be multiple, and in locations thought to be more specific for abuse (Wei, Stevens, Harrison, Mott, & Warner, 2012). Although it is clear that the smallest, earliest premature infants are at risk for osteopathy of prematurity, it is unclear at what gestational age prematurity is no longer significant for bone fragility, and at what age does past prematurity become no longer relevant in evaluating infant and childhood fractures. Differentiating fractures related to prematurity from abuse can be challenging. A recent review of chest radiographs in premature infants revealed rib fractures in approximately 2% of former premature infants, some of which were multiple and posterior in location (Lucas-Herald et al., 2012). In an effort to improve diagnostic certainty and reduce false allegations of abuse, further research into the relationship between prematurity and fracture susceptibility is needed.

Abusive Head Trauma

Abusive head trauma (AHT) is a leading cause of traumatic death and disability in infants and young children (Barlow, Thomson, Johnson, & Minns, 2005; Duhaime et al., 1992; Duhaime, Christian, Moss, & Seidl, 1996; Ewing-Cobbs et al., 1998; Ewing-Cobbs, Prasad, Kramer, & Landry, 1999; Hymel, Makoroff, Laskey, Conaway, & Blackman, 2007; Karandikar, Coles, Jayawant, & Kemp, 2004; Keenan, Runyan, Marshall, Nocera, & Merten, 2004). Early recognition is sometimes difficult (Jenny, Hymel, Ritzen, Reinert, & Hay, 1999; Laskey, Holsti, Runyan, & Socolar, 2004; Rubin, Christian, Bilaniuk, Zazyczny, & Durbin, 2003). Presenting signs can be non-specific; a caregiver history of head trauma is often absent, minimized or false; and external injuries are not universal.

Unfortunately, both over- and under-diagnosis of AHT can have adverse consequences. Over-diagnosis can exacerbate parental stress; increase hospital risk exposures, costs, and lengths of stay; and trigger serious, unintended, social and legal consequences. Under-diagnosis places children at risk for subsequent, more severe or fatal, inflicted injuries when returned to their abusive environments (Jenny et al., 1999). To further complicate these issues, multiple studies have shown that physicians demonstrate biases regarding child maltreatment; demonstrate an inconsistent response to severe child maltreatment; and struggle to define a reasonable degree of medical certainty of abuse (Benson, Swann, O'Toole, & Turbett, 1991; Jones et al., 2008; Lane & Dubowitz, 2007; Lane et al., 2002; Levi & Brown, 2005; Levi, Brown, & Erb, 2006; Lindberg, Lindsell, & Shapiro, 2008; Offer-Shechter, Tirosh, & Cohen, 2000; Rangel et al., 2009; Trokel, Waddimba, Griffith, & Sege, 2006; Wood et al., 2010).

In recent years, AHT has become a focus of intense public and legal scrutiny and debate (Barnes & Krasnokutsky, 2007; Donohoe, 2003; Gena, 2007, Tuerkheimer, 2009; Leestma, 2005; Lyons, 2003). Contested issues include: (1) the validity of the 'shaken baby syndrome' diagnosis, (2) the accuracy of clinical estimates of the timing of head injury, (3) our ability to reasonably and reliably exclude medical mimics of AHT; and (4) the 'reasonableness' or certainty of the diagnosis, in the absence of irrefutable evidence or a gold standard. Research is needed to help clinicians assess possible AHT more objectively and consistently. Success will require that clinicians and researchers adopt more evidence-based, probabilistic, and/or Bayesian approaches to AHT screening and diagnosis.

Data is emerging that the immature brain's pathophysiological response to traumatic brain injury (TBI) is different than that of the mature brain (Bittigau et al., 1999; Covey, Jiang, Alli, Yang, & Levison, 2011; Robertson, Soane, Siegel, & Fiskum, 2006). Optimal treatments for pediatric vs. adult TBI could differ. To improve clinical outcomes from AHT, novel therapies for pediatric TBI will need to be piloted in appropriate animal models.

To improve the focus and consistency of the medical response; to increase diagnostic specificity; to more accurately estimate the timing of head injury; to exclude medical mimics more reliably; to prepare for the testing of novel therapies; and to advance the protection and health of vulnerable children; the following are important research questions.

Which Clinical, Radiological and/or Laboratory Findings Should Trigger a Thorough Evaluation for Abuse in Young Children with Acute Head Trauma?

To improve the focus and consistency of the medical response, clinicians who treat acutely head-injured infants and young children need simple, reliable, highly sensitive, evidence-based, AHT screening tools that inform early decisions to either launch (or forego) an abuse workup. Research is also needed to further explore the feasibility, reliability and sensitivity of serum biomarkers that have shown promise as screening tests for brain injury and/or AHT (Berger et al., 2005, 2006).

Which Clinical, Radiological and/or Laboratory Findings have the Highest Specificity for Abuse in Young Children with Acute Head Trauma?

The societal responsibility to protect children from harm requires that cases of suspected child abuse be contested in civil and criminal courts. Although AHT has long been recognized as a clinically valid medical diagnosis (Narang, 2012), advocates for the 'falsely accused' have offered treatises challenging the validity of this diagnosis (Gena, 2007; Lyons,

2003; Tuerkheimer, 2009). To help clinicians respond optimally to these challenges, clinicians and researchers should apply evidence-based, probabilistic, and/or Bayesian approaches to diagnosing AHT.

What are the Specific Indications and the Appropriate Evaluations to Reasonably Exclude Medical Mimics of AHT?

To help confirm or exclude medical mimics of AHT, the forensic evaluation of head trauma would be enhanced by addressing the following questions:

- During infancy, can subdural hemorrhage(s) result solely from hypoxia-ischemia?
- Can asymptomatic, birth-related, subdural hematomas re-bleed or expand, spontaneously or from minor trauma, to trigger delayed, clinically significant, acute clinical deterioration and secondary brain injury?
- Can specific neuropathologic patterns or distributions of beta amyloid precursor protein (β-APP) expression be linked specifically to trauma, pure hypoxia-ischemia, or to infarction?

Which Clinical, Radiological and/or Laboratory Findings Best Define the Timing of a Closed Head Injury Event?

When medical professionals opine definitively and narrowly regarding the timing of a severe head injury event suspicious for abuse, a specific caregiver may become the focus of an investigation. Research is needed to more precisely define the onset and progression of those clinical, radiological, laboratory, and/or pathological findings that best facilitate precise estimation of the timing of pediatric head trauma.

Is Violent Shaking Sufficient—in Isolation—to Fully Explain the Acute and Long-Term Clinical Spectrum Described as the 'Shaken Baby Syndrome'?

When causality cannot be tested directly, most scientific disciplines apply the Bradford Hill criteria—a group of minimal conditions necessary to provide adequate evidence of a causal relationship between an incident and a consequence (Woodside & Davis, 2013). Researchers on both sides of the ongoing shaking debate should conduct research that facilitates application of the Bradford Hill criteria. Examples of research questions addressing each of these nine criteria include the following:

- **Strength of the association:** In young children who are violently shaken, is the relative risk of bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse significantly greater than the risk of these findings in children who experience a minor fall?
- **Consistency:** Has the association (between violent shaking, bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse) been repeatedly observed by different persons, in different places, circumstances and times?
- **Specificity of the association:** What percentage of young children who suffer closed head injuries resulting from a witnessed minor fall fail to reveal bilateral SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse?
- **Temporality:** Does violent shaking consistently precede the onset of new and acute clinical signs that led to medical evaluations and identification of bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse?
- **Biological gradient:** Is the frequency (or severity) of bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse higher in children who experience repetitive (or more prolonged) violent shaking vs. a single (brief) violent shaking event?
- **Plausibility:** Do other (animal, anthropomorphic, computer-simulated, and/or biomechanical) models or theories demonstrate a reasonable and consistent potential for violent shaking to trigger bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse?
- **Coherence:** What alternate, predominant, causal theories (for bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse) are contradicted specifically by the assertion of a causal relationship between violent shaking, bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse?
- **Experiment:** Do AHT prevention programs effectively reduce the incidence of AHT in the targeted population?
- **Analogy:** Would careful observations or study of mammalian predators shaking their prey to induce unconsciousness, or of young children restrained in car seats during rollover car accidents, provide supportive evidence of a causal relationship between violent shaking, bilateral diffuse SDH, extensive multilayered RH, diffuse brain HIE, and/or extracranial injuries independently concerning for abuse?

Research Related to the Testing of Novel Therapies for Pediatric TBI

Research should continue to develop and validate animal models that reliably replicate the primary injuries and secondary patho-physiological responses of the immature brain to traumatic injuries.

Medical and Related Child Maltreatment Systems Issues

Physicians working in the child maltreatment field necessarily collaborate with professionals in other disciplines (e.g., social work, mental health, child welfare, and law enforcement) – in a variety of settings and systems. Systems, such as hospital-based child protection teams (CPTs), child advocacy centers (CACs) and child fatality review (CFR) teams have been widely developed, based on extensive clinical experience but little experimental evidence. Some of these models are costly and may siphon funds from potentially preferable alternatives. Other models, such as CPTs are often poorly funded; empirical support of their effectiveness could well strengthen their request for better funding (Giardino, Montoya, & Leventhal, 2004). The National Association of Children's Hospitals and Related Institutions (NACHRI), now the Children's Hospital Alliance (CHA), has gathered substantial descriptive information on CPTs (National Association of Children's Hospitals & Related Institutions, 2013); notably missing are any research or evaluation findings. An important general question concerns the value of physician involvement in child welfare evaluations and management. This section focuses on physician involvement in multidisciplinary settings and systems concerning forensic evaluations of suspected maltreatment. There has been little research evaluating these systems; developing the knowledge base should help guide policy, practice and research.

Most children's hospitals have a CPT available for consultation when concerns of possible abuse or neglect arise (Kistin, Tien, Bauchner, Parker, & Leventhal, 2010; National Association of Children's Hospitals & Related Institutions, 2013). These teams vary considerably in how they are organized, who participates in them, and how they operate. Especially in larger institutions, their roles may include education, prevention and research, in addition to forensic evaluations of alleged maltreatment.

There are more than 750 CACs in the US, some located within children's hospitals (National Association of Children's Hospitals & Related Institutions, 2013). A CAC is a facility for the multidisciplinary investigation of alleged child maltreatment, mostly sexual abuse. The disciplines involved may include law enforcement, child protective services (CPS), prosecution, mental health, medicine, and victim advocacy. CACs aim to ensure quality and coordinated investigations, minimize further trauma to children, and to facilitate mental health interventions. CACs also vary in how they are organized, composed and operate, although accreditation through the National Children's Alliance has led to a degree of uniformity (Jackson, 2004; National Children's Alliance, 2013).

CFR teams review deaths where maltreatment is suspected, aiming to optimize the multidisciplinary investigation and to prevent future deaths. Members represent law enforcement, prosecution, CPS, medicine, mental health and some include community members (Hochstadt, 2006).

Are Hospital-based CPTs Effective?

There is a need to examine the impact of CPTs on child outcomes (e.g., recidivism), families (e.g., receipt of services), perpetrators (e.g., successful prosecution), and other professionals/hospital/public agencies (e.g., benefit to staff, policy and practice, cutting costs). Kistin and colleagues have developed a tool for measuring several of these aspects (Kistin, Tien, Leventhal, & Bauchner, 2011). One study found that a CPT helped prevent unnecessary CPS reports (Wallace, Makoroff, Malott, & Shapiro, 2007). A more refined probe could examine different models of CPTs and clarify what factors influence their effectiveness (Kistin et al., 2010). If found to be effective, analyzing cost-effectiveness will be useful.

Are CACs Effective?

There is a similar need to evaluate the effectiveness of CACs. Outcomes of interest include those pertaining to the child (e.g., number of interviews and examinations, referral for and receipt of mental health services), family (e.g., satisfaction with the investigation, referral for and receipt of services), perpetrator (e.g., successful prosecution) and professionals and agencies (e.g., confidence in the investigative process, cost). There is some evidence that CACs make the investigation easier for families (Jones, Cross, Walsh, & Simone, 2007). Here too, a more refined probe could examine different models of CACs and clarify what factors influence their effectiveness (Cross, Jones, Walsh, Simone, & Kolko, 2007). It may be especially interesting to compare those with and without physicians on site. One study found CACs made it more likely that children suspected of having been sexually abused were examined medically (Walsh, Cross, Jones, Simone, & Kolko, 2007). Another paper reported that contrary to what was expected, CACs did not reduce the number of forensic interviews (Moncada et al., 2008). They do appear to increase the likelihood of successful prosecution (Miller & Rubin, 2009). Notwithstanding this research, much remains to be done to answer the question posed over five years ago: Child advocacy centers: do they lead to positive case outcomes? (Faller & Palisci, 2007)

Are CFRs Effective?

Similar questions pertain regarding the effectiveness of CFRs. Outcomes of interest include: comprehensiveness and quality of the investigation, accuracy of the final determination, likelihood that siblings and other children in the household are evaluated, successful prosecution, changes in preventive policies or practices, and effects on maltreatment rates. Demonstrating fewer deaths due to maltreatment would be difficult given the low base rate. Research on CFR teams, however, would guide the development of these teams (Durfee, Parra, & Alexander, 2009) and delineate their role in preventing injuries and deaths (Onwuachi-Saunders, Forjuoh, West, & Brooks, 1999).

What Strategies Reduce the Barriers to Physicians Reporting Suspected Maltreatment to Public Agencies?

Physicians play an important gatekeeper role by identifying possible maltreatment and reporting to CPS and/or law enforcement. It has been amply established, however, that they often are reluctant to report, and children may continue to be maltreated (Jones et al., 2008). There is a need to better understand current barriers to reporting and more importantly to evaluate strategies to address them.

What is the Value of Physician Involvement in Child Welfare Evaluations and Management?

In addition to the models described above, child abuse pediatricians (CAPs) may directly consult to CPS, under a contractual arrangement. One study found that CAPs, with their greater training and experience, were able to render a definitive diagnosis regarding abuse in 42% of cases when physicians reporting to CPS had not (Anderst, Kellogg, & Jung, 2009). In almost half the remaining cases, CAPs had less concern for abuse than did the referring physicians. Such consulting arrangements remain relatively few and it would be useful to examine potential contributions by CAPs to the child welfare system.

Is there a Need for Federally Funded Research Collaborations?

For several decades, the National Cancer Institute (NCI) of the National Institutes of Health has supported national and international collaborations devoted to studying the causes of cancer in children. According to the NCI, approximately 10,400 U.S. children under the age of 15 were diagnosed with cancer in 2007 (American Cancer Society, 2007). In comparison, state and local child protective services (CPS) agencies estimated that 695,000 U.S. children were victims of child maltreatment in 2011 (U.S. Department of Health and Human Services, 2014). Like childhood cancer, AHT involves considerable morbidity and mortality (Barlow et al., 2005; Duhaime et al., 1992, 1996; Ewing-Cobbs et al., 1998, 1999; Hymel et al., 2007; Karandikar et al., 2004; Keenan et al., 2004) and other forms of child maltreatment (De Bellis, 2002; Felitti et al., 1998; De Bellis, 2001). Like childhood cancer, child maltreatment deserves a national infrastructure for collaborative, multicenter research that can provide sufficient data to answer our most important and pressing questions.

We have set forth what we think are important gaps in our knowledge concerning the medical forensic evaluation of suspected child maltreatment. Answering these questions should greatly advance knowledge and practice, and the protection of children. It is our hope that this article will be useful to funding agencies and organizations as well as researchers in the field.

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