Forensic pediatric radiology: studies in living and deceased children
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Citation for published version (APA):

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Summary, conclusion and future perspectives
SUMMARY

In this thesis we have brought together our studies describing the possibilities and impossibilities of establishing a diagnosis of child abuse in both living and deceased children, focusing on (forensic) pediatric radiology.

The thesis is divided into two parts, with the first part describing aspects of imaging in living children, suspected of being victims of child abuse, and the second part describing imaging in deceased children in a forensic setting.

In part I, chapter 1, we have given an overview of imaging techniques in suspected physical abuse, and the specificity of different types of fractures. Fractures are common in childhood; the likelihood of sustaining a fracture is 2.1% per year in children. Conventional radiography is the usual method for diagnosing fractures. A skeletal survey, according to international guidelines consisting of at least 20 radiographs, is the best way to detect fractures. The survey should be repeated 14 days after the initial trauma, to detect fractures that have formed callus over the period and were not visible at first examination. There is no consensus about the role of bone scintigraphy. Although it has a higher diagnostic yield in more anatomical complex locations, it is less sensitive for classic metaphyseal lesions (CMLs). CT should not be used as a replacement for conventional radiography to detect fractures, firstly because of the high radiation dose, and secondly because CMLs will go undetected. Whole-body short tau inverse recovery MRI (WB-STIR) has a low sensitivity for CMLs and rib fractures and should not be performed routinely. Certain fractures have a high specificity for abuse, but no fracture in itself is pathognomonic for child abuse. The injury should therefore always be related to the clinical history presented by the caregiver. Types of fractures most specific for abuse are rib fractures (especially posterior rib fractures) and CMLs. Dating fractures is not an exact science, but there is evidence that recent and old (healing) fractures can be distinguished.

In chapters 2 and 3 we reviewed the social-pediatric and radiological aspects of abusive head trauma (AHT). AHT is a relatively common cause of neurotrauma in young children, with an incidence of 14–40 cases per 100,000 children under the age of one year. About 15–23% of these children die shortly after the incident. One-third of the AHT survivors are severely disabled, one-third of them are moderately disabled and one-third have no or only mild symptoms. However, neurological problems can occur after a symptom-free interval and half of the AHT survivors have IQs below the tenth percentile. Clinical findings in AHT are dependent on the definitions used, but AHT should be considered in all young children with neurological signs and symptoms, especially if no or only mild trauma is described. Subdural hematomas (SDHs) are the most frequently reported finding. The only clinical feature that has been identified as discriminating AHT from accidental injury is apnea, with a positive predictive value (PPV) of 93%. A differential diagnosis of SDH exists, but the differential diagnosis of SDH in combination with fractures or other signs
of trauma is very limited. Detailed history-taking is the key to the diagnosis and should consist of a detailed history of the event that led to the clinical situation, medical history, growth curve, medical history of siblings, prior involvement of child welfare services and assessment of risk factors. In chapter 3 we reviewed the role of radiology in establishing the diagnosis of AHT. It is an important tool in describing the location and severity of the injury. Furthermore, it can help in the detection of abnormalities that can make the diagnosis more likely, such as rib fractures, or it can make the initial diagnosis less plausible, for example, if underlying bone disease is detected. Finally, it can help in assessing a prognosis, depending on the brain damage identified. In the acute phase, computed tomography (CT) is the best tool for neuroimaging. If CT is abnormal, early magnetic resonance imaging (MRI) including diffusion-weighted imaging (DWI) should be performed. The role of MRI, if initial CT is normal, is unclear as is the role of repeat CT if early MRI is performed. There is no evidence-based approach for the follow-up of AHT; both repeat CT and MRI are currently being used. A full skeletal survey, according to international guidelines, should always be performed to obtain information on possible underlying bone diseases, or injuries indicative of child abuse. Cranial ultrasonography is not indicated as a diagnostic modality for the evaluation of AHT. The final diagnosis of AHT can never be based on radiological findings only; it should always be made in a multidisciplinary team assessment, where all clinical and psychosocial information is combined and the diagnosis is established by a group of experts in the field.

In chapters 4 and 5 we studied all Dutch cases of AHT for which forensic medical expertise was requested by the Courts. The Courts request forensic medical expertise for 7.4 per 100,000 children per year in the Netherlands. Based on these forensic reports, risk factors for child abuse identified in other studies were found in this sample as well: prematurity, dysmaturity and twins/triplets were found in 27%, 23% and 10% of cases respectively, maternal age under 20 years in 17%. Of the parents, 60% had completed only primary or secondary education, and 38% of the families were known to child welfare authorities. It was striking that in 81% of families there was evidence of prior episodes of abuse. These results underline the need to improve the recognition of AHT by both child welfare authorities and the health care system. We also found that in 48% of the cases there was evidence of impact trauma. This resulted in chapter 5, where we tried to differentiate between impact and non-impact trauma based on radiological findings. We did not find any statistically significant differences in the distribution of radiological findings between the two groups, neither on neuroimaging nor on skeletal surveys. This indicates that, except in the case of a skull fracture, radiological findings cannot be used to determine the trauma mechanism leading to the injuries in AHT.

As SDHs are the most common manifestation of AHT, we performed a systematic review to identify the evidence for dating of SDHs on imaging findings, described in chapter 6. Age determination can be used to check whether there is a consistent history and can relate the injuries to possible perpetrators. We included 25 studies describing densities/ intensities
of SDHs on CT and/or MRI. Pooled time intervals showed a wide range and did overlap. Concluding, we did not find evidence that SDHs can be accurately dated on neuroimaging findings. In chapter 7 we described the knowledge and practice of Dutch radiologists regarding dating SDHs. We performed an online survey among neuroradiologists and pediatric radiologists. The results demonstrated that there is a considerable practice variation among Dutch radiologists regarding the age determination of SDHs. None of the participants was ‘very certain’ of their age determination. This implicates that dating of SDHs is not suitable for use in Court, as no uniformity among experts exists.

The first part of the thesis ends with chapter 8, a case report of a neonate with a CML of the distal right femur; a fracture of the long bones with a high specificity for child abuse. In this case it was detected after vaginal breech delivery, which we determined to be the cause of the fracture. With this case report we want to underline the need for a differential diagnosis, especially in children with fractures in whom abuse is suspected.

In the second part of the thesis, we describe imaging studies in deceased children. In chapter 9, we reviewed the current techniques used in postmortem imaging. As autopsy rates are declining, postmortem imaging could be an alternative. Postmortem radiology can be used in the clinical setting, in a forensic setting and to depict historical specimens, for example mummies. Conventional radiology has been in use for a long time, as the skeletal survey can be used to detect both fractures (in case of suspected non-natural death) and underlying bone disease or metabolic disorders. It is being used for fetuses and children up to four years of age; above this age it is not likely that additional information can be found with a skeletal survey. Furthermore it can be used to obtain detailed radiographs of specimens collected by the pathologist. Conventional angiography is upcoming; at the time of writing the review article, publications on the subject were scarce, but several papers have been published in the meantime. Information on ultrasonography is limited, but it can be used to guide biopsies in case of minimal invasive autopsy. Both CT and MRI are the most important modalities in current postmortem imaging.

In chapter 10 we described the normal cranial postmortem findings seen on postmortem CT (PMCT) in children who received a PMCT as part of a forensic examination because of a suspected non-natural death. We found that, although intravascular air is a normal postmortem finding, it appears more often in children in whom IV fluids have been administered.

In chapter 11 we described the diagnostic value of PMCT in this group, by addressing the correlation between cause of death diagnosed with PMCT and cause of death diagnosed with autopsy. PMCT and autopsy identified the same cause of death in 67% of the cases (kappa 0.5) using ICD-10 classification. PMCT did identify the majority of unnatural causes of death in children correctly (67%), but diagnostic accuracy in natural deaths in our study was low (0% agreement) compared to other publications. It does not give new insight into
the cause of death, if this is unexplained according to the pathologist, because PMCT failed to identify a cause of death in 98% of these cases; in the one case in which it did identify a cause of death which was not found with autopsy, this was considered to be a false positive finding. Our agreement of 67% is comparable with the agreement of 69% found in a MRI study, performed in the same age group. We conclude that, at present, PMCT cannot replace conventional autopsy in children.

In the ideal situation, PMCT is performed shortly after demise, to minimize the number of postmortem changes that can hinder interpretation of the images. In forensic practice, however, not all bodies are found shortly after their demise. Therefore, in chapter 12, we described the value of PMCT in neonaticide with delayed finding of the body, causing severe decomposition changes. In neonaticide, besides establishing a cause of death, an autopsy is also needed to determine gestational age and to assess possible live birth. In the case of late postmortem changes, PMCT is superior for estimating gestational age compared to foot length measurement with autopsy. There is no difference between PMCT and autopsy regarding the assessment of live birth or cause of death; both modalities were unable to determine these two parameters in any of the 12 cases in our sample. This might influence protocols for postmortem examination for suspected neonaticide in case of severe decomposition. PMCT is indispensable and should be performed compulsorily. In contrast to cases with little or no decomposition changes, described in chapter 11, a full autopsy might not always be necessary. We propose a new protocol for postmortem examination in the case of severe decomposition changes, consisting of a PMCT, visual inspection and sampling for DNA, toxicological and other forensic investigations.

The second part of the thesis ends with chapter 13, a case report describing pneumomediastinum and soft tissue emphysema after pediatric hanging. It illustrates one of the additional values of PMCT compared to autopsy, by demonstrating that the child was alive at the moment of hanging. Although cause of death was established with autopsy, these PMCT findings were not detected. Since they can be interpreted as vital signs at the moment of death, as the child must have been breathing to cause a pressure gradient that made the air escape, this information is of additional value to the other vital signs that were detected with autopsy.

CONCLUSION
Forensic pediatric radiology is a field that is developing rapidly. As establishing or missing a diagnosis of child abuse can have severe consequences, it is a field in which research should always be directed towards the diagnostic value of a test, and the amount of certainty that can be ascribed to each radiological report. In imaging in living children, many of these certainties have already been established. For the skeletal survey, for instance, the additional value of each of the radiographs is well known, as is the additional value of a repeat skeletal survey. But in forensic science, information on methods that are not
effective are also valued. For example, we found that the difference in injuries between impact and non-impact (presumed shaking) in AHT cannot be based on imaging findings. Furthermore, we did not find sufficient data to accurately date SDHs based on imaging findings, although in another study we found that dating SDHs is commonly practiced by radiologists. These results underline the need for evidence based reporting guidelines in forensic pediatric radiology.

Postmortem imaging is a new field; it is even referred to in some quarters as a new subspecialty, with many developments having taken place in the past few years. Although many advantages of postmortem imaging compared to autopsy are obvious, for example, the possibility of different observers reassessing the data over time, in our validation study the diagnostic accuracy is still low. The case mix seems to be an important predictor for the probability of agreement between PMCT and autopsy. In our study it performs better in non-natural deaths. In a prospective validation study on postmortem MRI in children, overall agreement was 56%, but this was raised to 89% if MRI was followed by minimal invasive autopsy. Here, age was an important predictor, as agreement for MRI and autopsy was 43% in fetuses aged 24 weeks or younger, 63% in fetuses older than 24 weeks and 69% in children. It is possible that the combination of CT, MRI and minimal invasive autopsy will enhance rates of agreement in the future, but at present PMCT cannot yet be seen as a replacement for a conventional autopsy. This is different in the subgroup of neonates with severe postmortem changes, because PMCT and autopsy are equally impeded by decomposition changes and the diagnostic accuracy of both modalities in determining the cause of death is low. In these cases, PMCT performs better in age determination of the neonates, making it an indispensable tool in the evaluation of neonaticide.

FUTURE PERSPECTIVES
During this PhD project, several developments have contributed and still are contributing to the professionalization of the field of forensic pediatric radiology. In 2012, the International Society of Forensic Radiology and Imaging (ISFRI) was founded in Switzerland. Its aim is to strengthen and develop the field of forensic radiology and imaging worldwide, including promoting best practice and developing international quality standards and guidelines in this field of imaging. A yearly congress is organized and since 2013 a scientific journal, aimed at forensic and postmortem imaging in particular, has been published: the Journal of Forensic Radiology and Imaging (JOFRI).

In the Netherlands, several national developments with opportunities to improve the forensic sciences are also in progress. In 2013, the Dutch Health Council published a report regarding the position of the forensic (health) sciences in the Netherlands. It was recommended that the forensic health sciences should acquire a more academic character and that an academic chair and accompanying research group should be established in order to develop a more evidence-based approach. Our radiological projects could contribute
to academic development within the forensic sciences. The Co van Ledden Hulsebosch Center, established in 2013, is an interdisciplinary center of expertise for forensic scientific and medical research. It is a collaboration between the faculties of physical sciences, mathematics and informatics, the Academic Medical Center and the Netherlands Forensic Institute. By collaboration with other disciplines, forensic questions that cannot be answered with forensic radiology, e.g. dating of SDHs, might be investigated with other techniques. For example, spectrometry, currently being used for the age determination of bruises, might also be a valid method of age determination of SDHs. In June 2012, a subsection on postmortem and forensic radiology was founded within the Dutch Radiological Society. Its aim is to bring together radiologists with an interest in post-mortem and forensic radiology in order to exchange experiences, develop imaging guidelines, and generate collaborative research.

From the side of pediatrics, the professionals with whom the radiologist works most closely in case of suspected abuse, several initiatives have been initiated. In the USA, child abuse pediatrics has been a new pediatric subspecialty since 2006. In the Netherlands this subspecialty does not exist, but several centers have acquired a great deal of expertise in diagnosing and managing child abuse cases. Currently, the Erasmus Medical Center Rotterdam, University Medical Center Utrecht and Academic Medical Center Amsterdam are working on a national center of expertise for child abuse (LECK), where colleagues can receive advice regarding diagnostics in and managing of child abuse cases. Furthermore, national guidelines and multicenter research projects will be developed by the LECK. Moreover, since 2011 a two-day course recognizing and responding to child maltreatment (the WOKK) is mandatory for residents in pediatrics.

An important theme in future forensic radiological child abuse research in living children is the dating of injuries. The more accurately injuries can be dated, the more precisely the history described by parents/caregivers can be verified. This question is valid not only for SDHs, but for fractures as well. In a meta-analysis, based on 82 fractures, a schedule with three age groups was constructed, but detailed fracture dating is still not possible. Other research fields might include the possibilities of advanced MRI sequences in neurotrauma (including spinal injuries) and the possibilities of whole-body MRI for the detection of fractures, potentially in combination with conventional radiographs of the joints to detect CMLs. Furthermore, in order to improve the detection of rib fractures, which are very specific for abuse but, especially if no callus is present, are easily missed on conventional radiographs, the value of ultra low-dose CT for the detection of rib fractures could be investigated.

Non-radiological topics in child abuse research are dictated by the fact that in many families child abuse is recurrent. As we, and others, found that a large proportion of AHT victims or their siblings were either known to social services, or had presented in a hospital with
complaints due to abuse, the major challenges in child abuse research are those in the field of early detection and effective interventions to stop abuse. Although these themes are not radiological, they can possibly reduce the number of children with ‘fractures in different stages of healing’. As the first (neurological) signs of AHT can be vague and do not always lead to neuroimaging, the studies of Berger et al. regarding serum biomarkers for pediatric brain injury are promising.43-48 If brain injury could be diagnosed with a routine laboratory test, this could be used as a simple screening instrument in the emergency department. Studies on effective interventions to prevent or stop abuse focus primarily on parenting programs. They mainly use derived outcome measures, such as parent-child interaction or mental health, and find variable results.49 A systematic review of behavioral interventions to stop child abuse found that risk assessment and behavioral interventions reduced child abuse in young children. Home visitation programs also reduced child abuse, but results were inconsistent.50

Future research on postmortem imaging in children should be aimed at determining the diagnostic and economic value of the combination of different techniques. Although neither PMCT nor postmortem MRI alone, are currently sufficient to determine the cause of death in children, the addition of minimal invasive autopsy with tissue sampling, postmortem ventilation and angiography could be promising.26,51,52 However, the combination of these techniques will probably be introduced at a very high cost. Efficiency should be taken into account when introducing a new method. Furthermore, knowledge regarding normal postmortem findings is needed, both to understand pathological changes and to develop a timescale for postmortem radiological alterations. It is possible that the question will arise of who will interpret the postmortem imaging studies. As it requires specific knowledge of both postmortem changes and the interpretation of imaging findings, either a pathologist with an interest in radiology or a radiologist with an interest in pathology could be suitable candidates.29 Different methods are practiced in different centers. Regardless of the direction in which postmortem imaging evolves; forensic pediatric radiology will continue to play an important role in the diagnosis of child abuse, in both living and deceased children.
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