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Comparison of postmortem ultrasound and X-Ray with autopsy in fetal death: Retrospective study of 169 cases ☆

Olivier Prodhomme *, Catherine Baud, Magali Saguintaah, Nancy Béchard-Sevette, Julie Bolivar, Stéphanie David, Ikram Taleb-Arrada, Alain Couture

Department of Pediatric Radiology, Arnaud de Villeneuve Hospital, Montpellier, France

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ABSTRACT

Objective: To compare the diagnostic yields of joint analysis of ultrasound and X-Ray compared to autopsy in the setting of fetal death.
Material and methods: Retrospective study of postmortem fetal ultrasounds performed between January 2009 and December 2013 in addition to X-Ray in the work-up for cases of fetal death. Inclusion criteria were a complete anatomical ultrasound study and the availability the foetopathology report. Cases with autopsy refusals were excluded. When cases of TOP were included, antenatal imaging data (US, CT and/or MRI) were available. All scans were performed by a senior pediatric radiologist.
Results: We collected 169 patients. The gestational age was 27 weeks ±6 days [15–38 WG]. The population consisted mainly of fetuses originating from TOP (164/169 [97%]). Only 5 cases involved in utero fetal deaths (IUFD). Half (50%) of the conditions involved were cerebral. Others were polymalformative syndromes (20%), kidney diseases (10%) and miscellaneous (20%). The duration of the exam was about 10–15 min. Complete concordance between the findings of postmortem imaging and autopsy was observed in 81% [137/169] of cases.
Conclusion: Ultrasound allows a comprehensive post-mortem study complementary to standard X-Rays. In fetal deaths situations, US is much more relevant than in any other postmortem conditions. Ultrasound, although less effective than MRI, shows a benefit/drawback balance that proves very interesting, especially in the youngest fetuses. Besides it is more available and realistic to use it in a systematic practice.

1. Introduction

Foetopathology encompasses two distinct situations. The first one is unexpected in utero death (IUD) at any stage of pregnancy, whether or not a cause was suspected on the basis of clinical data and/or prenatal imaging. The second one is medical termination of pregnancy (TOP) which is allowed in France until the last day of pregnancy. In the first situation the goal is to try to establish the cause of death to offer adequate prevention for a future pregnancy. In the second situation, the aim is to achieve the comprehensive assessment of fetal malformations in order, ultimately, to improve prenatal counseling as far as a risk of recurrence can involve a future pregnancy.

Results from early studies showed agreement between prenatal ultrasound and postnatal examination in 40–60% of cases [1,2]. A specific comparison of post-mortem examination and prenatal ultrasound has shown similar rates of concordance, with post-mortem providing major additional information in 30–40% of cases [3,4]. In this study, post-mortem confirmed the ultrasound findings in 98% ($n=46$) of cases. This demonstrated complete agreement between prenatal ultrasound and post-mortem findings in 22 cases (46.8%), and major agreement in 24 cases (51.1%). Post-mortem provided significant additional information in 27.7% of cases, and defined the diagnosis in 12.8% of cases. In a more recent study [5] performed over a 12-month period, 153 prenatal ultrasound cases were identified, of which 47 were also examined by autopsy and were thus suitable for comparison in this study (30.7%). Complete agreement between ultrasound and post-mortem findings was found in 22 cases (46.8%). In 24 cases (51.1%), major agreement between ultrasound and post-mortem findings was seen. Of these, 11 had minor additional findings at post-mortem examination and 13 were found to have significant additional findings at post-mortem (27.7%), adding to the eventual diagnosis. In 12.8% of cases, post-mortem examination provided a definitive diagnosis. Only in one case was there complete discordance in the findings of the ultrasound examination and the autopsy (2.1%). No major disagreement between ultrasound and post-mortem findings was seen in 98% of cases. These studies confirm the accuracy of

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* Correspondence to: Department of Pediatric Radiology Arnaud de Villeneuve Hospital 371, avenue du Doyen Gaston Giraud 34295 Montpellier Cedex 5 FRANCE. Tel.: +33 467336017; fax: +33 467336018.
E-mail address: o-prodhomme@chu-montpellier.fr (O. Prodhomme).

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Table 1
US findings.

US findings	
Brain 84/169 (49%)	**Callosal a(dys)genesis 19 (11.2%)** Brain malformation (Dandy–Walker) induced by spinal meningoencephalocele ventricular (V) dilatation: uni-V 2, bi-V 8 and tri-V 7 holoprosencephaly 8, cranial meningo-encephalocele 5,brain calcifications 3, ventricular hemorrhage 3 ***N*=2:** rhombencephalosynapsis, schisencephaly, Dandy–Walker malformation, inter-hemispheric cyst, septal agenesis ***N*=1:** ventriculitis, pachygyria, sub-ependymal nodules, torcular thrombosis, parenchymal cyst, choroid plexus cyst, vermian agenesis, anomaly of brain echogenicity, brain tumor
Kidney 37/179 (22%)	**Uni- or bilateral kidney(s) agenesis 7 *N*=5**: posterior urethra valves, bilateral poly-cystic nephromegaly, uni- or bilateral mlulticystic kidney(s), horse-kidneys **Others**: kidney hypoplasia 3, nephromegaly without cyst 2, renal duplicity, isolated kidney cyst 1
Spinal cord 18/179 (10.7%)	Myelomeningocele 12, abnormal position of the conus medullaris 6
Congenital diaphragmatic hernia 9/179 (5.3%)	Confirmation of prenatal finding 8, new finding 1
Others 24/179 (14.2%)	Lung hypoplasia 6, microphtalmy 4, ascitis 3, pleural effusion 2, heart myomas 2, ano-rectal malformation and cloacal malformation 2, cystic adenoid malformation of the lung 2, hepatic cyst 1, epiphyseal calcification 1, hip dislocation 1

Table 2
X-Ray findings.

X-Ray findings	
Normal	69/179 (41%)
Major findings 63/179 (37%)	**Severe skeletal dysplasia 15 (9%)** including osteogenesis imperfecta 2, Apert syndrome 1, hypophosphatasia 1 **Spinal malformation included dysraphism 15 (10.7%)** Mandibular hypoplasia and other facial anomalies 9, semi-circular canals agenesia 4, cranial meningoencephalocele 4, polydactyly 3, radial agenesis 3, epiphiseal calcifications 2, limb hypoplasia 2, enterolithiasis 2, other 4
Minor findings 37/179 (22%)	Brachymesophalangy/brachymetacarpy 9/3, clubfoot 8, macro/microcephaly 6/3, nasal bone agenesis 4, arthrogryposis 3, hip dislocation 1

Table 3
All discrepancies between US postmortem examination and autopsy (all false negative or incomplete findings of US study).

Discrepancies 32/169 [19%]
Heart malformations (9) Cerebral malformation (10): hamartoma (3), heterotopia (3), rhombencephalosynapsis, hemangiopericitoma, lissencephaly, cerebellar maturation delay, hypoplasia
Other: esophageal atresia (2), ano-rectal malformation (4), cystic adenoid malformation (1), bilateral cataract (2), tracheal atresia in complex malformation (1), polycystic kidney (1), hepato-renal recessive polycystic kidney (1), congenital diaphragmatic hernia (1)

prenatal ultrasound diagnosis and emphasize the continuing importance of post-mortem examination to refine and, in some cases, define the diagnosis. However, the accuracy of postmortem ultrasound has never been studied, which was the objective of this work. Up to now, only five publications about the use of postmortem US were found and only two concerning the topic of fetal death [6–10]. In the older one published in 1989, US was perform only when autopsy was refused and therefore do not compare their results with the gold standard [9]. The second one, published in 2014, presents the experience of postmortem US in only 2 macerated fetuses immersed in a water bath [6]. The aim of the study is to determine the accordance rate between postmortem US examination (combined with X-Ray) and autopsy (gold standard).

2. Material and methods

We performed a retrospective study of postmortem fetal ultrasounds performed between January 2009 and December 2013 in addition to X-Ray in the work-up for cases of fetal death. Inclusion criteria were a complete anatomical ultrasound study and the availability the foetopathology report. Autopsies were performed by a dedicated foetopathologist. Anatomopathology (brain excepted) was performed by an experimented pathologist trained fetal medicine. Brain studies were performed by a pathologist specialized in neuropathology. Cases with autopsy refusals were excluded.

All patients had X-Ray (babygrams) anteroposterior and lateral views. Data from X-Rays were analyzed as follows: maturation, mineralization, evidence of focal dysostosis or bone dysplasia, soft tissue study.

Some fetuses (9/169, 5.3%) also had postmortem fetal MRI and one had also a post-mortem CT (0.6%), but they were not taken into account in assessing the value of postmortem ultrasound.

When cases of TOP were included, antenatal imaging data (US, CT and/or MRI) were available when performing the ultrasound scans. In all case but one prenatal US was performed (99.4%). Prenatal MRI was performed in 51/169 (30.2%) and CT scan in only 2 cases (1.2%).

Ultrasound studies were performed on a Philips iU22 system (Philips Healthcare, Eindhoven, The Netherlands). Three probes were used: 8–5 MHz micro-convex probe (mainly for brain study), 12–5 MHz linear probe (for almost all other explorations) and 17–5 MHz probe especially for the youngest fetuses (< 18–20 WG) and superficial explorations. The fetus was placed in the supine position to explore brain, eyes, chest, abdomen and pelvis. Then the fetus was placed in the prone position to study the posterior fossa through suboccipital window and to study the spinal cord, vertebrae and kidneys. All scans were performed by a senior pediatric radiologist. The following organs were systematically examined: brain, posterior fossa, spinal cord, eyes, thymus, lungs and pleura, diaphragm, abdominal solid organs, digestive and urinary tract. Since the study was retrospective, body temperature could not be collected. The primary endpoint was agreement, or not, between the results of postmortem imaging (joint analysis of