Amniotic fluid embolism: Then and now

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Abstract

Background: The first case report to describe amniotic fluid embolism that appeared in 1926 in *Basil-Medico* is translated from Portuguese to English.

Case: A patient with a dead fetus for several weeks, presented in labor and died suddenly with fetal squames evident in the maternal pulmonary vasculature at autopsy.

Conclusion: As can be seen from the translation, this case report is remarkably similar in many of its features to some of the eight patients described 15 years later in the first English language discussion of the disease by Steiner and Luschbaugh in *JAMA*. An enigma presented by this first case remains today: fetal material in the maternal pulmonary vasculature appears specific for amniotic fluid embolism at autopsy but not in living patients.

Keywords

Maternal mortality, maternal–fetal medicine, perinatal medicine, complications, intensive care medicine

Introduction

A recent study of the maternal uterine vasculature in 57 women undergoing emergency peri-partum hysterectomy found intravascular fetal material in 19, while only 1 woman was diagnosed with amniotic fluid embolism (AFE) on the basis of her clinical course.¹ Yet, fetal material in the maternal pulmonary vasculature at autopsy is the only laboratory finding that is widely considered to be diagnostic of AFE.² Indeed, the definition of AFE used by the British incorporates this finding as part of their diagnostic criteria.³ Even so, there is growing evidence that fetal material in the maternal vasculature of living women is not diagnostic of AFE, and indeed may be present without causing direct harm. While the logical conclusion is that the disease state might be directly related to the amount of fetal material entering the maternal circulation, maternal autopsies do not seem to support this. To date, there have been no instances reported in the medical literature in which fetal material has been found in the maternal pulmonary vasculature in women dying of causes other than AFE although there are unpublished, anecdotal observations of pathologists observing fetal material in the lungs of parturients dying from causes other than AFE (personal communication).² The specificity of fetal material in the maternal pulmonary vasculature in death—but not in life—does not make much sense, but it is what the available data demonstrate.

In this context, it is instructive to look back at the historical record in which the disease was first described. The two papers published in *JAMA* in 1941 by Steiner and Luschbaugh are the most widely cited, although some authors assign the first description of AFE as a distinct entity to JR Meyer who published a single case report in 1926 in the Portuguese language in *Brazil Medico*.^{4,5} It appears that Steiner and Luschbaugh were unaware of this case report when they published their case series of eight patients, 15 years later. With papers published in multiple languages, and before the advent of computerized indices, it is hard to be sure that JR Meyer's case was the first. However, an Irish physician, Lois D Courtney asserted that this was the first case report in his 1974 review paper, and we have not come across any evidence to refute this.⁶ Steiner and Luschbaugh did review the literature across several languages in 1941 and did not find any other case reports, either.

Although case reports are the weakest sort of evidence for studying disease, there are remarkable similarities between the first descriptions of AFE by physicians on different continents who spoke different languages. For this reason, there is merit in taking another look at these first two papers on AFE—and in particular, providing an English

translation of JR Meyer's work to make it accessible to a wider audience. As a footnote, most of the citations that we have seen cite the paper improperly as it appeared in volume 40, issue 2—not volume 2 as commonly cited.

The Brazil Medico case report

Translation: JR Meyer. Embolia pulmonar amnio-caseosa. *Brazil-Medico* 1926; 40 (2): 301–303

“Pulmonary embolism by amniotic vernix caseosa”

Introduction

In the present work, our aim is to consider a type of embolism not recorded in the literature and which is characteristic of late pregnancy, and which occurs as a result of breaks between the amniotic membranes and maternal venous circulation.

Case report

MC, 21 years old, multiparous, comes to the clinic in the eighth month of pregnancy, and in apparent early labor. Vaginal examination reveals: cervix dilated to just over two fingers, the integrity of the bag of waters, and a dead fetus possibly in the left occiput anterior position.

After the membranes rupture, there is a moderate uterine discharge that, coincident with a small thermal elevation, is slowly becoming more abundant and thicker, until it acquired a frankly purulent character, 3 days later. The thermometer indicated a temperature of 38.8°C and pulse of 144 beats per minute.

After the rigors of intense and sharp sweating, both the pulse and the temperature returned to normal. Labor, which had ceased to raise

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the temperature, began again 15 days after breaking the bag of waters and ends spontaneously in about 25 min.

After experiencing severe dyspnea, seizures, and slight, but continuous bleeding, only diminished by the use of uterine tamponade after the delivery of the afterbirth, during the expulsion of the fetus, the patient died, suddenly attacked with frank cyanosis. The patient's body was then submitted for pathologic examination.

Pathological examination

Macroscopic maternal injury: The only data worth mentioning found during the autopsy, were slight congestion of the lung parenchyma and massive dilatation of the right retro-ovarian venous plexus. The uterus internally presents above the placental insertion area of the upper third of its posterior wall, a yellowish and rough area.

Macroscopic fetal injury: In addition to severe maceration, cadaveric fetal examination shows copious amounts of purulent material within the trachea, bronchi, and bronchioles.

Macroscopic placenta injury: This soon drew attention, at first sight, the almost complete disappearance of the membranes, which are only represented by a small area of uniform appearance, measuring about 4×2 cm. Both the umbilical cord and the fetal surface have a yellowish color. They lost their normal transparency. We cannot highlight or detect remnants of the amniotic sheet.

Microscopic maternal injury: For the histological study, there are several interesting lesions in the lung. In many places, the blood capillaries are congested and they exhibit one or other syncytial cell, permeated by the red cells that fill them. Elsewhere, numerous neutrophil leukocytes can be seen, preferably occupying the small venous vessels of the alveolar septa. Different and particularly curious are the structures that are inside the capillaries, through careful analysis. In general they are elongated and tortuous, as may be seen in Figure 1 [in the original article], suggesting at first small portions of fibrin, yet distinguished by fibrinous formations at its most consistent and because they do not give color reactions of this substance. Within a pulmonary capillary formation is seen a long and transparent, consisting of a hair shaft of fetal lanugo. The remaining viscera have little to deserve mention. Excluding the uterus, where are perceived areas with signs of postpartum endometritis, it can be said that the other organs are normal.

Fetal microscopic injury: Represented by large numbers of neutrophils and some mononuclear, is noted an intense pneumonic process not only in the alveoli but also within bronchioles.

Microscopic lesions of the placenta: Histological sections of the placenta, including a piece of the membranes that we described, show a remarkable marked leukocyte infiltration of the connective tissue inter-amnion-chorion. The research of germs by direct methods reveals Gram-positive organisms only on the surface of the endometrium, placenta, and fetus in the lung parenchyma. The maternal blood cultures do not reveal nothing abnormal.

Discussion

Given the history and the lesions found in this case, one might think that, in addition to bleeding from placental delivery, one of the factors responsible for the death has been the infectious process that developed rupture of amniotic membranes.

However, alongside such other occurrences not yet known figure in obstetrics, which seems endowed with practical interest, because it may be the cause of sudden death occurring during the delivery or in late gestation.

We refer to a type of embolism determined by the epidermal cells of the fetus. In this case, the elongated structures that we describe in the capillaries of the lung, with its characters absolutely equal to those elements, prove this possibility.

As in adults, the superficial layers of fetal epidermis, usually suffer a process of scaling that, in this case, mixing with sebum, gives rise to the vernix caseosa.

These generally form lumps that remain adherent to the integument of the fetus that are more or less immovable. These particles, however, are not always stationary and, as evidence that can be carried from one point to another, there is the fact that they are found very often, both in the sediment of centrifuged amnion, as in the meconium or in the lungs of fetuses asphyxiated in uterus.

In the presence of such data (and how easy it is to verify by examination of Figures 2 and 3 [in the original article] represent, respectively, these elements in a state of peeling skin and in a lung of a fetus suffered intrauterine asphyxia), it is reasonable to admit that they are capable of penetrating the maternal organism.

Thinking this way, we found such structures in the middle of the lung parenchyma, as we were looking for the supposed route of these formations. We also had the opportunity to find them not only in the veins of the endometrium but also the retro-ovarian venous plexus.

Given such information, seems pretty clear that there is a special form of embolism that is not described until now, we refer to the specification of amnio-caseosa.

Another important fact that explains, in our case, the existence of such a process is the phenomenon of dissolution of the chorion, as it can be seen in the simple macroscopic examination of the placenta.

Whether or not caused by the action of bacteria, once the continuity between the amniotic membranes and the uterine wall is disrupted, its access to the uterine contents directly with the inner surface of the uterus becomes possible.

At a later stage, with placental abruption, the entry of amniotic fluid remaining in the veins occurs. With the negative pressure of the same or effect of uterine contractions, the likely penetration of epidermal cells in the maternal venous circulation becomes possible, leading to the occurrence of the process embolic mentioned.

From what can be deduced from this pathology, the same type of embolism can occur as a result of high rupture of membranes, trauma, or lesions of the uterus which result in the amniotic fluid contact with the deciduous veins.

From this we see the attention it deserves, both in pathology and in practice, consideration of the venous system of the uterus—means of access to numerous morbid agents that affect the pregnant body.

We register our thanks to Prof. Cunha Motta, dismissed by the kind cooperation in preparing this work, drawing the pictures that accompany it.

Comment

The single most striking aspect of JR Meyer's paper is that he found fetal material in the maternal pulmonary vasculature and was able to find some of this same material in the ovarian veins. The case occurred in a woman who presented with ruptured membranes, fever, and a dead fetus. She subsequently went into labor 15 days later. After a 25-minute labor, she developed respiratory distress, convulsions, and intractable bleeding and then died abruptly. At autopsy, she had evidence of chorioamnionitis in addition to the fetal material in her lungs.

Fifteen years later, and on a different continent, Steiner and Luschbaugh published their case series of eight maternal deaths in the United States (Chicago, Illinois). They also found abundant fetal material in the maternal pulmonary vasculature. In their series, three of the women entered labor with dead fetuses and one of these cases echoed the JR Meyer's case, as she also had sepsis. Similarly, in one of the patients, Steiner and Luschbaugh found "amniotic fluid emboli" both within the lungs and in the uterine veins. What is often over-looked in the subsequent literature is that these pathologists did acquire a control group of 34

parturients dying in the third trimester of other causes. They did not find any fetal material in the lungs of anyone without AFE with the one exception of finding a small amount of mucin in the pulmonary vasculature of a single patient.

While intravascular fetal material in the living does not seem to be specific for AFE, autopsy findings may be a different story. The fact that pathologist on two continents both gave the first descriptions of this disease on the basis of finding fetal material in the maternal pulmonary vasculature lends credibility to the specificity of this finding for AFE, at least in fatal cases.

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Declaration of conflicting interests

None declared.

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Ethical approval

Patient consent not required as no current case is discussed.

Note

1. We were unable to obtain the artwork to reproduce the figures in the original article, so we have not included the figures and captions in this translation. These can be seen in the original article.

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