

Classification of perinatal death

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SUMMARY Three paediatric pathologists, one perinatal paediatrician, one obstetrician, and one epidemiologist separately used information collected on 239 babies in an attempt to validate the Wigglesworth classification of perinatal deaths. This was first done using clinical data only, then using the combination of clinical and gross necropsy findings and finally using clinical, gross necropsy, histological and any other information (for example, chromosome analyses, microbiological investigations). Only 14 (6%) of deaths changed groups within the Wigglesworth classification when gross necropsy findings were considered as well as clinical findings, and altogether only 21 (9%) changed classification when complete investigations were available. There was an unacceptable amount (15%) of disagreement between the classifiers, largely the result of failure to comply with the rules laid down for classification. We set out amendments to Wigglesworth's original definitions to clarify certain ambiguities.

The number of classification systems of the causes of perinatal death have increased over the years. Obstetricians^{1 2} have derived classifications that differ substantially from those of pathologists.³⁻⁷ Paediatricians tend either to quote basic statistics,⁸ or to use classifications derived by pathologists rather than obstetricians.^{6 7} Despite the multiplicity of classifications, and firm but divergent opinions about which may be best, there has been little assessment of the repeatability or the usefulness of any of them.

The aim of any classification must be to derive strategies to understand the reasons for, and ultimately prevent, perinatal mortality. Wigglesworth argued that this was the only valid reason for a classification.⁵ He suggested that any classification should be as simple as possible and presented in such a way that the results give clear indications of priorities for prevention, and that the classification should be used to indicate the areas of health care provision most in need of alteration. He put forward a simple classification in which he allotted deaths to one of only five categories. It was devised so that it could be applied with reservation to cases where necropsy investigation had not been undertaken, although he acknowledged that necropsy would permit the most accurate categorisation.

The simplicity of the classification is most attractive

and the fact that it can be used when necropsy has not been undertaken is helpful. Nevertheless, problems were encountered when it was used in one specialised unit⁹ and other problems were encountered by one of us (JWK) in attempting to apply it to a regional study of perinatal mortality with a high necropsy rate.

The present study was set up with three objectives: firstly, to define more accurately the groups of the classification, particularly in respect of problems already encountered; secondly, to investigate the extent to which classification of cases was changed by necropsy; and thirdly, to assess the reproducibility of the classification when undertaken by people with an interest in perinatal medicine and pathology but with different professional backgrounds.

Method

The six authors (three perinatal pathologists, one perinatal paediatrician, one obstetrician, and one epidemiologist) met first to discuss problems already identified in the Wigglesworth classification and to define its groups more clearly. At a subsequent meeting each member of the group was given the clinical, gross necropsy findings, and details of further investigations of 233 perinatal deaths that had been collected as part of a multicentre study of

perinatal deaths. Each member of the group was asked to use the Wigglesworth classification and classify the death firstly on the clinical information alone, secondly on the clinical information together with the findings from the macroscopic examination at necropsy (which included the dissection of organs), and thirdly to classify the death taking account not only of the clinical and necropsy findings but also the histological and microbiological examinations, and chromosome analyses.

Each member of the team carried out the classification without knowledge of the classification made by the other members. After 89 perinatal deaths had been classified, the team met to discuss problems encountered and to compare results of the initial classification exercise. At this stage there was substantial disagreement. For each case over which there was disagreement, extensive discussion took place to try and define more clearly the rules of the classification. Once these rules had been delineated more clearly, the team was presented with a further 144 cases to classify. Before this, the obstetrician (IM) had classified the 233 deaths using the Aberdeen classification.²

The following analyses of the data were carried out: (i) identification of cases where there was still disagreement concerning the appropriate Wigglesworth grouping; (ii) a study of all cases where necropsy and other results had made a difference to the classification; (iii) a comparison of the classifications made by all six observers of the last 144 cases (to give an estimate of interobserver reliability); and (iv) a comparison of the Aberdeen classification with the Wigglesworth classification, after taking into account all necropsy and other findings.

Results

CLARIFICATION OF THE CLASSIFICATION

Group 1

The original Wigglesworth group 1 was described as 'normally formed macerated stillbirth'. After discussion the group was renamed 'deaths before the start of labour'. In the absence of other evidence, the presence of maceration should be taken to indicate that death preceded the onset of labour. Antepartum fetal deaths with lethal malformations were excluded, as were the miscellaneous disorders (see group 5).

Much discussion concerned the distinction between lethal and potentially lethal malformations and minor malformations. It was suggested that macerated infants with minor isolated lesions (such as a small ventricular septal defect) should remain in this group, but that any infants with multiple minor anomalies (for example, cleft lip and a skeletal

defect) should be classified in group 2. Because many antepartum fetal deaths are associated with abruption, it was thought that a subgroup might be appropriate.

Group 2

Group 2 was originally described as 'congenital malformations (stillbirth or neonatal death)'. Infants with minor or potentially treatable minor malformations should not be included here unless they formed part of a complex of at least two malformations and they had died before the start of labour. Deformations—that is abnormalities of form secondary to a fetal disease or functional impairment—could also be included here. Thus pulmonary hypoplasia that follows oligohydramnios may be included, although we thought that this difficult diagnosis should always be supported by the presence of other deformations or malformations.

Group 3

Group 3 was described as 'conditions associated with immaturity (neonatal deaths only)'. It was decided that this group should be titled 'conditions associated with preterm birth or immaturity' and include only livebirths of under 37 weeks' gestation.

Infants weighing less than 1000 g should be presumed to belong to this group irrespective of the time of death. Larger preterm infants are likely to have suffered from birth asphyxia if they die at less than 4 hours of age. Thus any infant dying at less than 4 hours, delivered preterm, and weighing more than 1000 g, should be coded to group 4 below unless a specific condition was present.

Neonatal deaths with infection, even congenital infection, should be included here if they were delivered preterm, although specific infections—for example, group B streptococcus and toxoplasma, other viruses, rubella, cytomegalovirus, and herpes virus—should be coded group 5. The epidemiologist's view was that if some infections remained in group 3, then all infections should stay in group 3. The counter argument was that 'important and interesting' infections should go to group 5.

We came across problems with infants delivered at full term but who had disorders normally associated with prematurity such as hyaline membrane disease, intraventricular haemorrhage, and necrotising enterocolitis. It was decided that these should be coded as group 5.

Group 4

Group 4 in the original classification comprised 'asphyxial conditions developing in labour (fresh stillbirth/neonatal death)'. After discussion it was decided that this group should include all fetal

deaths of whatever weight without malformations or specific disorders, provided that fetal death occurred during labour. In the absence of other information all fresh stillbirths should be included in this group.

Liveborn infants weighing over 1000 g who died at less than 4 hours of age should be included in this group. If the fetal death occurred during an intervention such as a caesarean section, in the absence of labour, the case should be classed group 4, as should any infants surviving longer than four hours for whom there was evidence of cerebral birth trauma or asphyxia. Massive antepartum haemorrhage such as abruption could form a clear subgroup.

Group 5

Group 5 was originally described as 'specific conditions other than above; and should include the following: blood group incompatibilities, inborn errors of metabolism, twin to twin transfusion syndrome, hydrops not associated with malformation, specific or unusual infections, conditions usually associated with preterm delivery but occurring in a baby born at full term, tumours, hamartomas, neonatal deaths of term babies that are totally unexplained, fetomaternal bleeds, or anything completely out of the ordinary.

The decision tree evolving from the refined classification is shown in the figure.

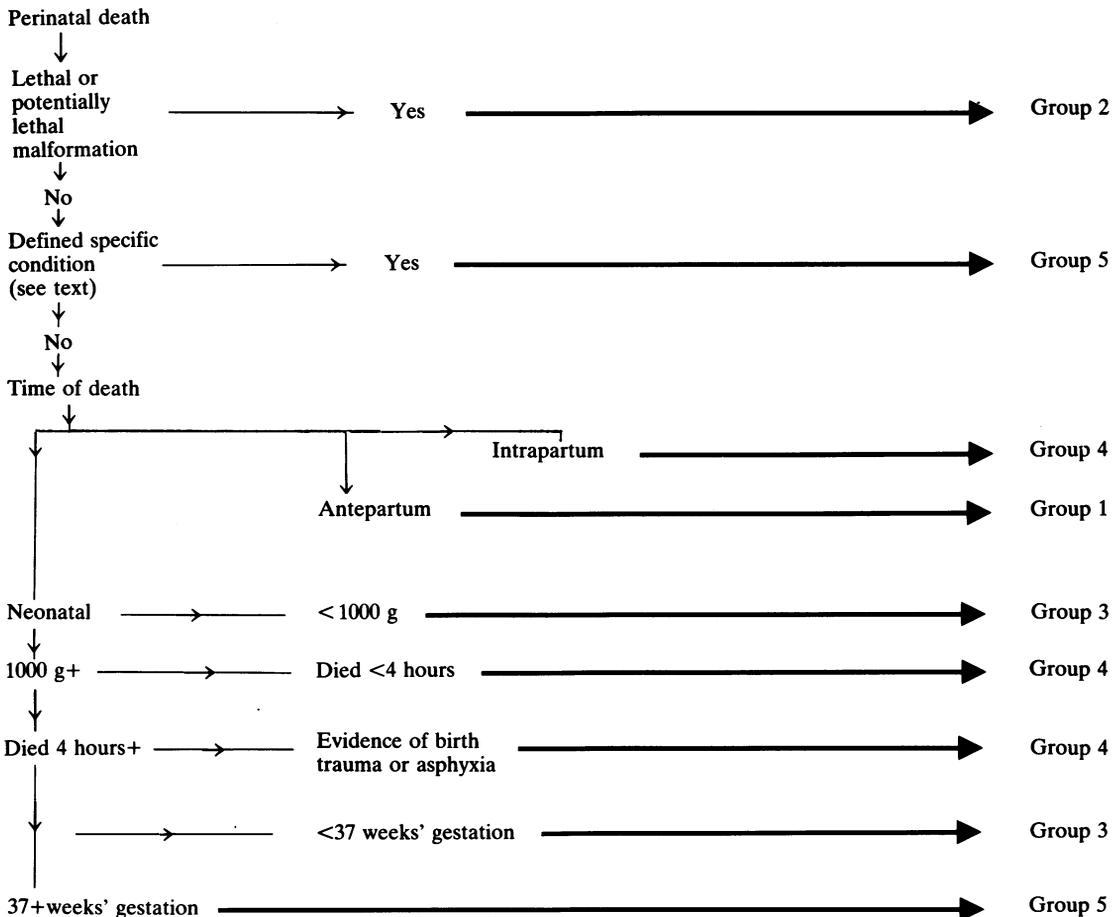


Figure Decision tree for the revised Wigglesworth classification.

Table 1 Changes in Wigglesworth classification after necropsy and other findings had been reviewed

Case No	Classification			Comment
	Clinical only	Clinical plus necropsy	Clinical, plus necropsy, plus other findings	
10	3	4	4	Tentorial tear
25	3	2	2	Congenital heart disease
117	3	2	2	Congenital heart disease
184	3	2	2	Congenital heart disease
65	1	2	2	Congenital heart disease
196	4	2	2	Congenital heart disease
123	1	2	2	Gastrointestinal defect
143	1	2	2	Congenital heart disease
137	2	5	5	Clinical congenital heart disease; at necropsy idiopathic arterial calcification
155	2	5	5	Congenital heart disease—collapsed during catheterisation; classified to iatrogenic
181	1	5	5	Idiopathic arterial calcification
151	4	4	5	Stormy labour, weighed 3580 g at 37 weeks, died at 7 hours. β haemolytic streptococcus grown
204	4	5	5	Weighed 3640 g at 40 weeks; died at 2 days of pneumonia
231	2	5	5	Antenatal scan showed cystic mass in neck; at necropsy non-rhesus hydrops found
175	4	5	4	At necropsy signs of sepsis found; on histological examination no sign of sepsis
97	3	3	5	β haemolytic streptococcus grown
100	1	1	5	Congenital cocksackie A virus isolated
145	1	1	5	Listeriosis
290	3	3	2	Trisomy 21
193	3	3	5	β haemolytic streptococcus grown
244	3	3	5	Enzyme defect

CHANGES IN THE CLASSIFICATION AFTER NECROPSY

Of the 239 cases considered, 14 (6%) were reclassified once the gross necropsy findings were considered (table 1) and it can be seen that most were because of the discovery of congenital anomalies at necropsy. In addition, consideration of other data including results of histological and microbiological examination further changed eight cases including one that had changed between the clinical to the gross necropsy classification (case 175). Interestingly, this case reverted to the classification that had been based on clinical grounds. Four of the other cases resulted from the identification of specific infections, one was an enzyme defect, and one a chromosome anomaly. Thus in all, 21 (8%) cases were reclassified as a result of necropsy and other findings. It is unlikely that 8% of diagnoses would be changed in geographic populations because the sample that we studied was not a representative series of perinatal deaths. It emanated from teaching hospitals with high rates of referral of complicated cases, and could therefore be expected to have a higher than normal proportion of unusual findings.

INTEROBSERVER RELIABILITY

The first sample of 89 cases were used as a learning exercise. The observers then compared results on the subsequent 144 cases in order to assess reproducibility. In all, for each set of case notes, there were 15 possible pairs of results. For each of the three assessments (clinical only, clinical together with gross necropsy, and clinical and all findings) there were $144 \times 15 \times 3$ —that is 6480 possible pairs.

Disagreement at initial classification was found for 963 pairs (15%). It should be noted that the 85% agreement rate is the interobserver variation and does not measure the accuracy of the classification. Major causes of disagreement concerned (i) whether a fetal death had actually occurred during labour, (ii) the class to which an immature infant should go, and (iii) how intrauterine infections should be classified. Despite agreement beforehand about definitions, clinical experience tended to override them.

PROBLEM CASES

Of the 233 cases considered there were 10 in which

there was considerable discussion before eventual agreement. These are discussed below as examples of the most difficult cases to classify.

Case 16 was a diabetic cherub with visceromegaly who died before the onset of labour. The problem was whether this should be group 1 or 5; the ultimate decision was group 5.

Case 39 was a stillbirth that died before the onset of labour. Clinically it was recognised as having abnormal palmar creases and prominent heels, possibly as a result of a chromosome abnormality. At necropsy a solitary horseshoe kidney was found. The chromosome culture failed. This was eventually classified as group 2.

Case 51 was an infant born by caesarean section for severe pre-eclampsia at 26 weeks. The baby weighed 510 g and died aged 1 day. There was hydatiform change in the placenta. Should this be group 3 or 5? As this is an unexpected and unusual finding it was put into group 5.

Case 53: liquor had been draining for weeks since amniocentesis. The baby was born at 28 weeks' gestation, weighed 1160 g, and died at 90 minutes. Necropsy and histological examination confirmed a congenital infection but no specific organism was isolated. Various members of the team wanted to put it in group 3, 4, or 5. The rules, however, are clear and it was put into group 4.

Case 72 was an infant delivered at 28 weeks' gestation (1200 g) who died at 5 days. This was a breech extraction and the baby was clinically recognised as having aortic thrombosis with gangrene of the right toes and a flaccid left leg. Permission for a full necropsy was refused and external examination only was carried out. There was discussion about whether this should be classified to group 3, 4, or 5, but it was finally put in group 5.

Case 77 was a breech extraction at 27 weeks weighing 1271 g which died at 20 minutes. There had been prolonged rupture of the membranes with oligohydramnios and the infant had hypoplastic lungs. The rules put this into group 4.

Case 170 was born at 35 weeks' gestation weighing 1588 g. He had minor anomalies of the hands, limbs, and skull and died of massive pulmonary haemorrhage and disseminated intravascular coagulation at 7 days; this was put in group 3.

Case 186 was born at 37 weeks' gestation weighing 2330 g, and died at 4 hours of age. At necropsy the baby was found to have a polycystic right kidney, bilateral hydronephrosis and a double right ureter, and hypoplastic lungs. Gross necropsy examination showed a subarachnoid haemorrhage, hyaline membrane disease, and pneumonia. Further investigations identified mycoplasma pneumonia. The question was whether this should be classified as group 2, 3, 4, or 5. The team was divided, but the rules indicated group 5.

Case 212 was born at 36 weeks' gestation weighing 2240 g. It was dead at birth with mild hydrocephalus and undescended testes. Clinical notes stated clearly that death was before labour. Labour was of unknown duration. According to the rules this should have been coded as group 1 unless there was doubt as to whether the death occurred before or during labour.

Case 160 was born at 38 weeks' gestation weighing 2200 g and died at 4 days. There was cardiac malposition, a single umbilical artery, undescended testes, and anomalous pulmonary segments. The baby died of a massive pulmonary haemorrhage with a subarachnoid haemorrhage. There was much disagreement as to how this should be classified, but the final decision was that it should be group 5.

Table 2 Aberdeen clinicopathological, and amended Wigglesworth, classifications after taking account of clinical and necropsy findings

Aberdeen classification	Wigglesworth classification							Total
	1(a)	1(b)	2	3	4(a)	4(b)	5	
Premature, cause unknown	22	0	4	49	4	0	7	86
Mature, cause unknown	11	0	1	0	9	0	1	22
Mechanical	0	0	2	3	8	0	0	13
Toxaemia	4	0	1	10	2	0	1	18
Antepartum haemorrhage	0	11	0	6	0	7	2	26
Malformation	0	0	43	0	0	0	3	46
Maternal disease	5	0	0	1	0	0	1	7
Rhesus incompatibility	0	0	0	0	0	0	6	6
Miscellaneous	0	0	0	0	0	0	4	4
Infection	0	0	0	1	0	0	4	5
Total	42	11	51	70	23	7	29	233

Subgroups (a) without and (b) with a history of abruption.

RELATION TO OBSTETRIC CLASSIFICATION

The way in which the Wigglesworth classification relates to the Aberdeen classification is shown in table 2. It is clear that the two are measuring different things. For example, of the 86 deaths classified by the Aberdeen rules as 'premature cause unknown' the Wigglesworth classification would distinguish the 22 dying before labour from the four dying of intrapartum causes, and the 11 dying of malformations or specific disorders from the 49 dying of disorders associated with immaturity.

Conversely, of the 30 that by the Wigglesworth classification would have been coded as intrapartum asphyxia, the Aberdeen classification would have distinguished the four 'premature—cause unknown' and the nine 'mature—cause unknown' from eight 'trauma' and two 'toxaemia'.

Discussion

As different observers become more aware of the usefulness of classifying perinatal deaths to compare performance over time and between centres, it is of importance to ensure that the classifications used are repeatable. A recent publication showed that the Aberdeen obstetric classification, which had been used for the Scottish National Survey and the Northern Regional Health Authority Survey, allowed differences in its interpretation.¹⁰ These differences had developed both north and south of the border during the years since its introduction. The classification has been in use for 30 years and only in 1986 was a revised classification published with attention paid to current problems such as the death of the very immature neonate.² The authors concluded that different assessors can classify a series of deaths in a similar way, provided that close attention is paid to the definitions and working rules. They found that different groups of clinicians reached similar conclusions in 97% of cases submitted for assessment as long as they had a copy of the rules before them.

Nevertheless, there is considerable debate about the interpretation of all classifications of perinatal deaths. As most classifications used in the United Kingdom are based either on the Aberdeen or Wigglesworth classifications, we will confine our discussion to these two. Table 2 shows that, in general, the two classifications measure different things. Which is the more useful? Proponents of the Aberdeen classification suggest that it is important to know how many infants died from toxaemia, yet using that classification one is unlikely to find this out from the way the data are presented. The classification defines this group as having a diastolic pressure of 90 mm Hg or more on two separate days after 28 weeks' gestation together with appreciable

proteinuria, and in the absence of hypertensive disease before pregnancy (which is comparatively common in pregnant women). Is it reasonable to assume that any normally formed baby born to a woman with this condition and dying prematurely is the result of toxaemia? Prevention of pre-eclampsia in the mothers may not have prevented most of these deaths. Similarly, the Aberdeen classification of 'trauma' (or under the revised classification, 'mechanical causes'), covered deaths from birth trauma and antepartum asphyxia associated with problems in labour such as disproportion, malpresentation, cord prolapse, cord compression, or breech delivery in babies of 1000 g or more. If there is no clinical history of difficulty in labour, but necropsy evidence of trauma is present, the deaths are not classified as 'mechanical causes' but as 'unexplained'. In other words, the trauma group does not indicate the number of deaths with evidence of birth trauma, but is a count of the number of deaths with abnormal presentation or method of delivery. It assumes that malpresentation is accompanied by asphyxia and is thus responsible for death. Malpresentation is, however, common in small babies, so this group might just be an indication of the proportion of preterm infants.

On the other hand, the Wigglesworth classification does not, in its primary analysis, examine causes of death in individual cases, so that subclassification within the primary groups may be necessary. The classification relies heavily on gestational age and time of death, and Wigglesworth has suggested that a number of assumptions can be made from these. A large number of deaths in group 1 (antepartum stillbirths) suggests that more attention to antenatal care might be warranted; many deaths in group 2 (congenital malformations) perhaps indicates that more attention should be given to preconceptional care; high numbers in group 3 (deaths associated with immaturity) might suggest that changes in neonatal intensive care were warranted; a high proportion of deaths in group 4 (intrapartum asphyxia) might indicate that obstetric care could be improved. It is always anticipated that group 5 will be a small group of miscellaneous causes from which few generalisations could be made, although it is a crude measure of the standard of perinatal histopathology, and other investigative services.

Since this study was carried out there has been much discussion among the participants. Each still prefers the classification that he has either developed or grown up with, but a majority were in favour of the revised Wigglesworth classification—perhaps because half the observers were perinatal pathologists. At the moment the topic of classification,

especially the difference between the Aberdeen and the Wigglesworth classifications, is largely a British phenomenon. Nevertheless, publications from many countries are now appearing using the Wigglesworth classification, interpreting the results to emphasise areas in which medical care could be improved.¹¹⁻¹³

Classification should be precisely defined so that it is easily applied by different people. In 10 cases (4%) no consensus was reached. This is too high, but most of these cases could have been classified by strict adherence to the revised rules.

Changes in classification occurred in 8% of cases when information derived from the necropsy and the results of all investigations were added to the clinical information. Though necropsy investigations contribute to the accuracy of classification, the extent of change is modest and is certainly acceptable when the alternative is omission of deaths not coming to necropsy. In most population studies this will be in excess of 30%.

Interobserver variability was 15%, and this degree of disagreement is too high for most studies. Scrutiny of the results showed that many discrepancies resulted from one person failing to apply the rules correctly, while 220 (23%) of the pairs of disagreement concerned the 10 cases that were resolved after further discussion.

We must reflect on the cause of the discrepancies. Is the classification insufficiently precise for universal application, or were the classifiers at fault? It is likely that the answer is the second possibility. When classification is undertaken by those with previous experience in a particular area it is likely that they will be swayed by their experience rather than adhering to guide lines, however precise. Possibly the answer would be to have the coding done by trained clerks rather than doctors, although there are cases (as we have seen) in which considerable judgment is required and it would be advisable

in these doubtful cases to have a consensus decision from a medical panel.

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