

Epidural haemorrhage of the cervical spinal cord: a *post-mortem* artefact?

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Spinal epidural haemorrhage is a rare entity that occurs uncommonly in adults and rarely in children. It has a typical clinical presentation, although to date, the cause for the majority of cases remains unknown. We present a series of cases where epidural haemorrhage was identified at *post-mortem*, principally to the cervical cord, in cases outside the age range usually reported for clinical epidural haemorrhage, and with no underlying pathology to account for the finding. We present a hypothesis for a *post-*

mortem cause for this finding and consider that, in the absence of any other identifiable causation, then this is a *post-mortem* occurrence similar to that of the Prinsloo–Gordon artefact of the soft tissues of the neck. This finding must be interpreted with care so as not to make the mistaken diagnosis of a nonaccidental head injury based on its finding, especially in the absence of intracranial, cranial nerve, optic nerve or eye pathologies.

Keywords: cervical spine, epidural haemorrhage, *post-mortem* artefact, trauma

Introduction

Since Caffey first described the shaken baby/shaken impact syndrome in 1972 [1], many papers have been written on the subject putting forward several theories as to the possible mechanism of injury involved to produce the classical findings observed at *post-mortem* [2,3]. Initially the emphasis of these theories centred upon the forces and mechanisms involved to produce the intracranial and optic pathologies, but more recently the effect of excessive hyperflexion or hyperextension of the cranio-cervical junction has been questioned [4,5]. With these more recent hypotheses has come the need for a more detailed examination of the cervical aspect of the spinal cord, which may previously have received less detailed examination than the brain, optic nerves and eyes. How-

ever, in those dying of so-called 'sudden infant death syndrome' (SIDS) or in cases with no clinical history of head and/or neck trauma, this area may receive little, if any attention.

The spinal cord may be removed attached to or independent of the brain either using an anterior or posterior (or combined) approach [6]. In cases of suspected infant trauma a posterior approach is preferred to allow for a formal posterior neck dissection to examine for soft tissue and muscular injuries caused by hyper-extension/flexion of the neck. One of the observations that may be seen during this procedure is the presence of epidural haemorrhage to the cervical aspect of the spinal cord, an observation reported to occur infrequently in adults and with few publications in infants [7–37].

Although there are both medical and therapeutic causes of epidural haemorrhage in infants some relate this rare occurrence to be suspicious and supportive of nonaccidental head injury as is hypothesized to occur in shaken baby syndrome. However, it has been our obser-

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vation that this phenomenon is, in fact, a common occurrence at *post-mortem*, occurring in those infants who die with no history of head or neck injury at all. We propose that in the absence of a demonstrable natural disease, therapeutic procedure or history of *antemortem* trauma, that this may be purely a *post-mortem* artefact similar to the Prinsloo–Gordon artefact seen to the soft tissue structures of the neck [38]. We present a small series of cases to illustrate this occurrence in children who died in the absence of *antemortem* head and/or neck trauma. These cases show that this may be a previously undescribed *post-mortem* artefact, which must not be misdiagnosed as attributed to hyperflexion/hyperextension of the neck. A small control series of known cases of head and neck trauma are also reported to illustrate the different findings to the neck area in such cases. Finally, two cases of children with evidence of *antemortem* physical abuse are presented who died in the absence of intracranial, optic nerve or eye pathology yet had evidence of cervical epidural haemorrhage. These latter cases are presented to illustrate that this finding should not be taken on its own to diagnose an *antemortem* 'shaking' episode prior to death.

Case reports

Six cases are presented to illustrate the position and magnitude of the epidural haemorrhage observed in children dying with no history or *post-mortem* evidence of head or neck injury. These cases are selected from the forensic paediatric autopsies undertaken over a 9-month study period to best illustrate the location and appearance of this *post-mortem* finding for the purposes of this report. During this period 14 forensic paediatric autopsies were undertaken using the double-doctor system (G.N.R. and C.J.H.P.). Of these, 12 were nontrauma cases of which six showed this phenomenon.

All of these cases were examined at the same mortuary, utilizing a double-doctor approach, i.e. a paediatric (C.J.H.P.) and forensic pathologist (G.N.R.) undertaking a joint examination with production of a joint *post-mortem* report. On all occasions the neck was opened as the first invasive *post-mortem* procedure from a posterior approach. Prior to the opening of the neck a cervical puncture had been performed to remove a sample of cerebral spinal fluid (CSF) for the purpose of microbiology/virology examination, toxicology and metabolic studies. This was performed without a skin incision, with the nee-

dle inserted immediately below the level of the foramen magnum, that is, at the level of the cranio-cervical junction. A layered posterior muscle dissection was performed, noting any soft tissue or muscular injuries, followed by the opening of the entire spinal column, which proceeded from the lumbar area going towards the head. The findings illustrated were observed immediately upon removal of the cervical portion of the spinal column. In each case, the brain and spinal cord were examined by a single forensic neuropathologist (W.S.).

A control series of two cases where the child died following nonaccidental head and/or neck trauma and two cases where the child had been abused but died of causes other than that of a head injury are also presented. The *post-mortem* procedures of these cases were in the same manner as detailed above (cases 7 and 8 G.N.R. with paediatric review of *post-mortem* report and histology, cases 9 and 10 C.J.H.P. and G.N.R.). Neuropathology was undertaken on the brain and spinal cord of all four cases (cases 7 and 8 another, 9 and 10 W.S.). These cases are selected to illustrate the presence or absence of this phenomenon in relation to children dying of nonaccidental injury and abuse. They allow a hypothesis to be presented within the discussion as to why this haemorrhage may or may not occur under the circumstances of the cases presented.

Nontrauma, nonabuse cases [Figures 1–5]

Case 1 A 15-month-old male child was found dead at home in a cot. There was no history of *antemortem* trauma. When the child was discovered dead the father was seen to carry it out of the house with the neck unsupported and possibly 'shake' it in an attempt to resuscitate it. The cause of death was found to be attributed to methadone toxicity (child weight 10.5 kg, blood methadone 298 ng/ml). At *post-mortem* he was found to have an area of epidural haemorrhage to the cervical cord, 1.0 cm long, 2.0 cm below the foramen magnum (FM), a second 0.5 cm lateral area 4.5 cm below the FM and a third 0.8 cm long 6 cm below the FM. There were no brain, optic nerve, eye, posterior neck muscle or spinal nerve root injuries present.

Case 2 A 13-day-old female infant was found dead in bed with her parents. There was no history of *antemortem* trauma. At *post-mortem* she was found to have congenital

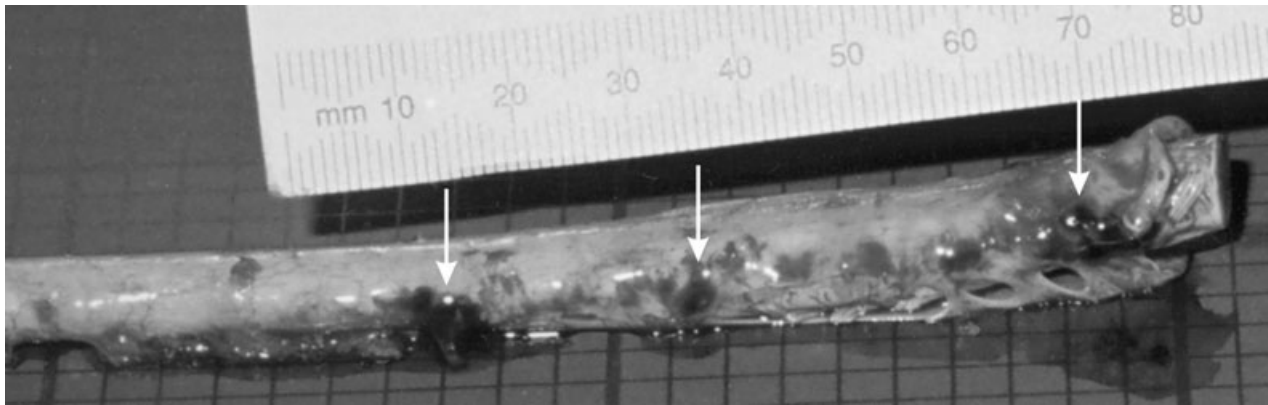


Figure 1. Posterior view of the cervical and upper thoracic cord showing three areas of lateral epidural haemorrhage (arrows) associated with the spinal roots (arrows) in a case with no history of head or neck trauma or physical abuse.

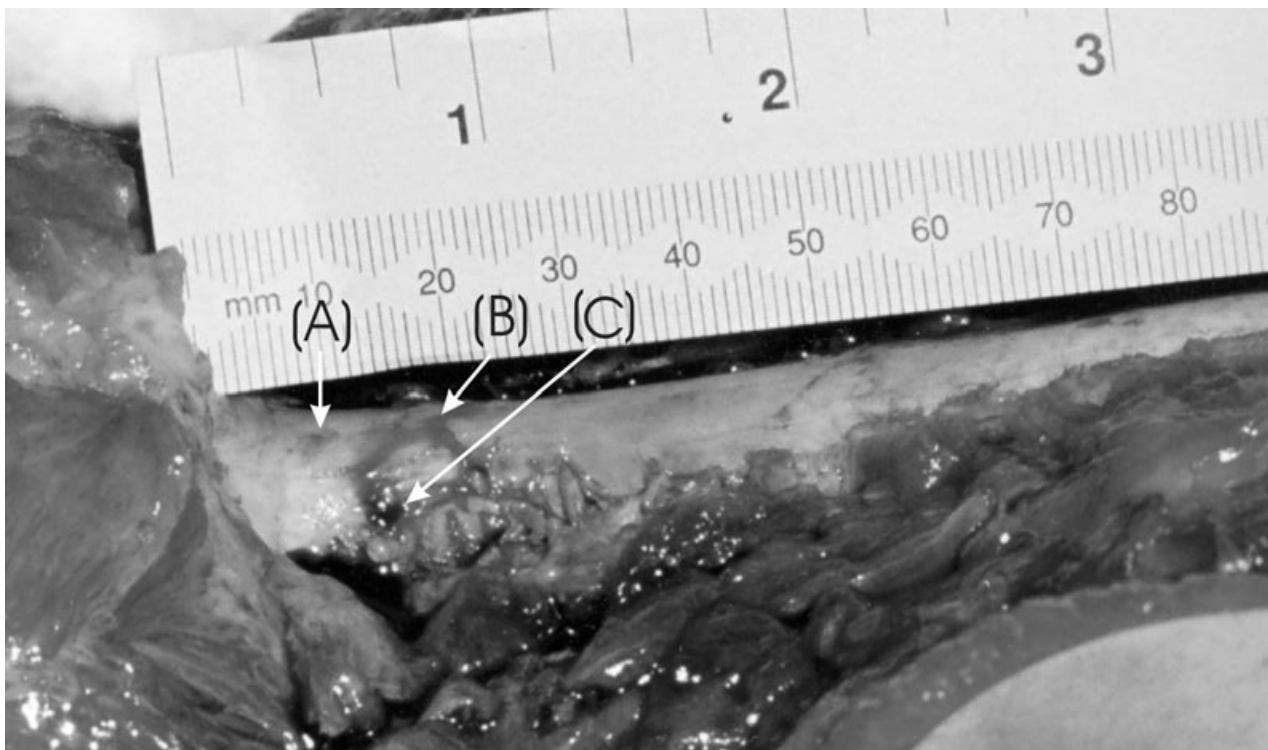


Figure 2. Posterior view of the cervical cord *in situ*. (A) Site of cerebral spinal fluid tap at *post-mortem*. (B) Thin diffuse area of epidural haemorrhage. (C) Lateral epidural haemorrhage associated with nerve root in a case with no history of head or neck trauma or physical abuse.

heart disease (patent fossa ovale, equal-sized left and right ventricles and patent ductus arteriosus). Diffuse posterior-lateral epidural haemorrhage was found to the cervical spinal cord extending intermittently to other parts of the cord, with prominence to the thoracic aspect.

Case 3 A 2-month-old male child was found dead at home in bed, clothed where he had been left unsupervised.

There was no history of *antetrauma*. At *post-mortem*, a thin layer of epidural haemorrhage was seen to the posterior aspect of the cervical spinal cord.

Case 4 A 3-month-old male child was found dead at home in his travel cot at the base of the parent's bed where he had been placed the previous night when the parents had retired to bed. There was no history of *antemortem*

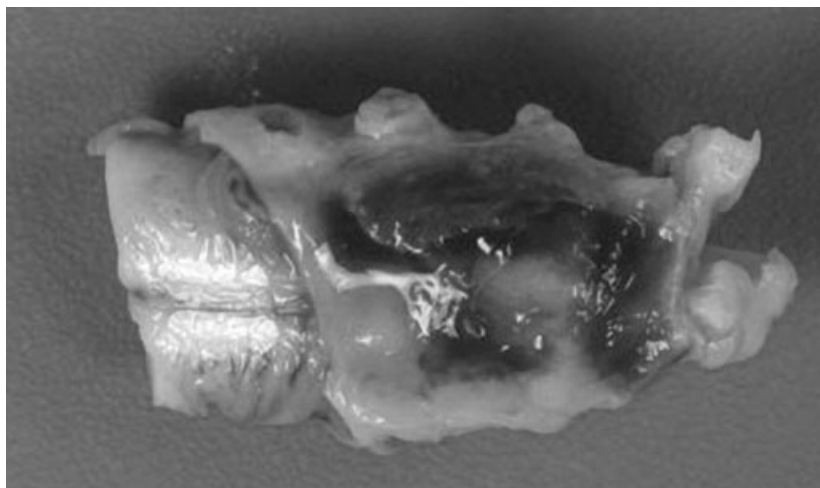


Figure 3. Postfixation portion of upper cervical cord (posterior view) showing diffuse epidural haemorrhage in a case with no history of head or neck trauma or physical abuse.

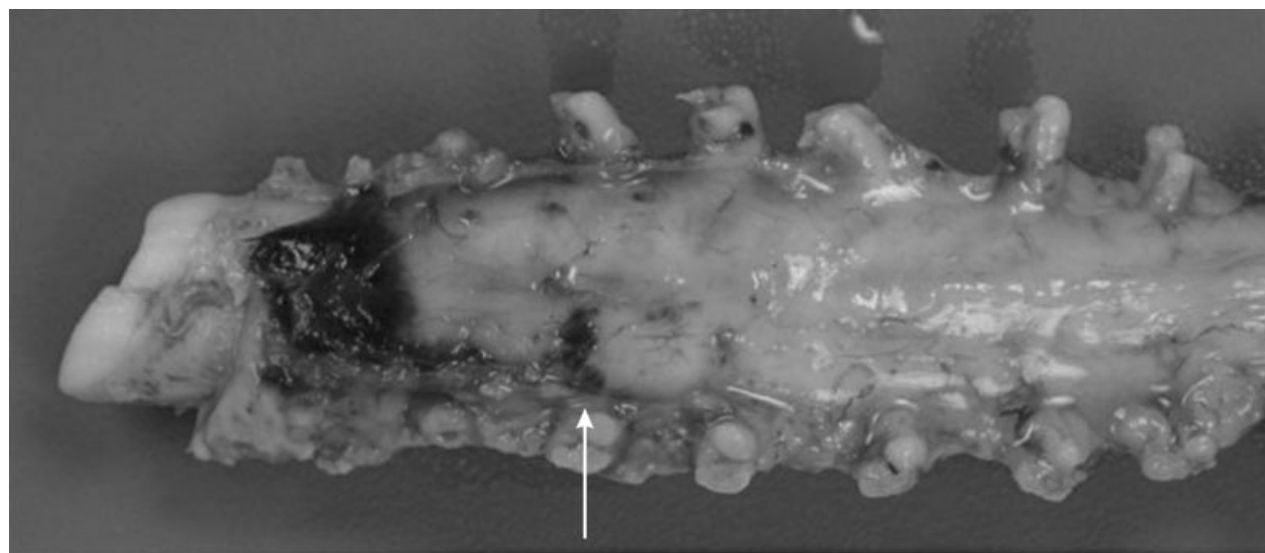


Figure 4. Postfixation portion of upper cervical cord (posterior view) showing diffuse epidural haemorrhage and separate lateral area of haemorrhage (arrow) associated with nerve root in a case with no history of head or neck trauma or physical abuse.

trauma. At *post-mortem*, a 0.8 cm diameter area of fresh epidural blood was found to the posterior aspect of the upper cervical cord.

Case 5 A 14-month-old male child was found hanging upside down, dead, wedged between the radiator and the bed in which he had been placed. There was no history of *antemortem* trauma. At *post-mortem*, epidural haemorrhage was found to the right and left lateral aspects of the cervical spinal cord at the level of C1. Epidural haem-

orrhage was also seen to the lumbar area of the spinal cord.

Case 6 A 5-week-old male child was with his mother on a settee. The mother fell asleep only to awake later to find the child dead with its face close to, but not resting, on the cushions at the head end of the settee. There was no history of *antemortem* trauma. On this occasion, both epidural and subdural haemorrhage was seen to the cervical spine.

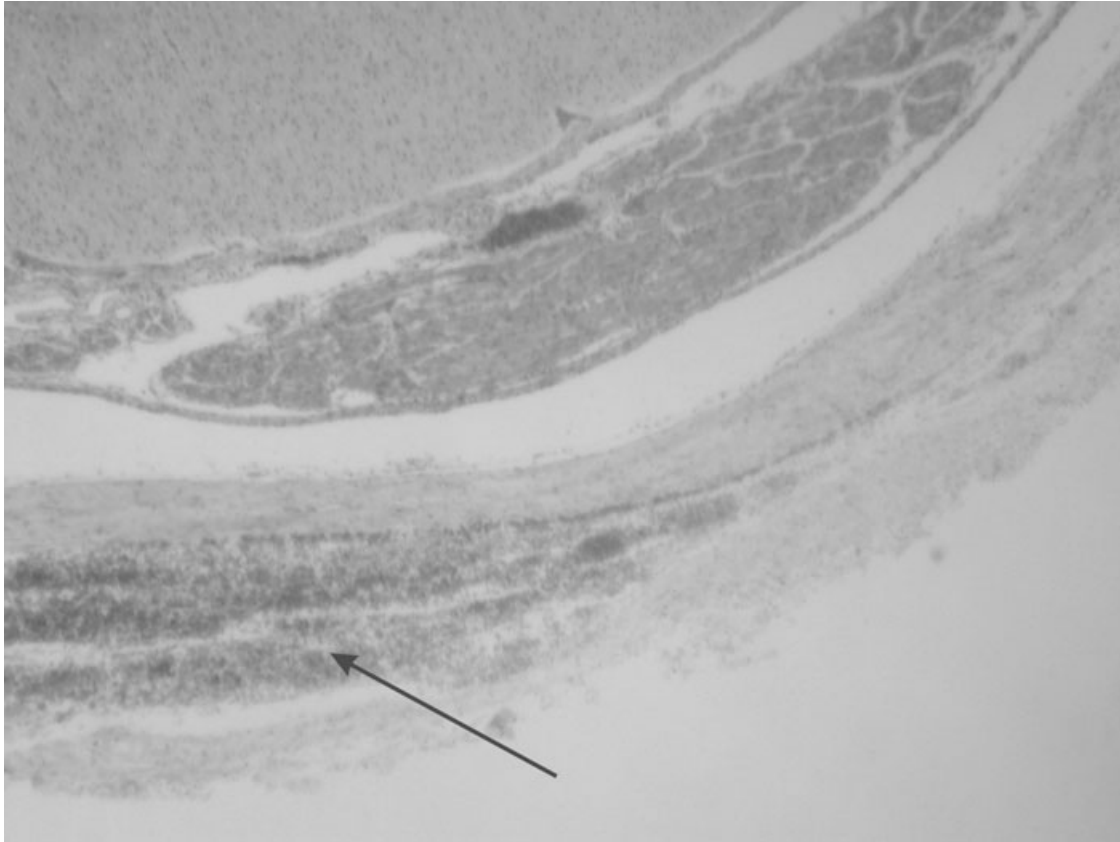


Figure 5. Microscopic haematoxylin and eosin stained section of fresh epidural haemorrhage (arrow) in a case with no history of head or neck trauma or physical abuse.

Nontrauma, abuse cases [Figure 6]

Abuse case 7 A 16-month-old female child was found to have been the victim of repeated episodes of abuse. However, her principle pathology at *post-mortem* was an empyema, pneumonitis and myocarditis. A prominent area of posterior epidural haemorrhage was found to the posterior mid cervical aspect of the spinal cord. There was, however, no evidence of intracranial, optic nerve or retinal pathology and no soft tissue neck injuries to support a traumatic causation.

Abuse case 8 A 13-month-old male child was found to have been the victim of repeated episodes of abuse. At *post-mortem* the principle findings were severe fresh lower thoracic and abdominal injuries. A thin layer of fresh epidural haemorrhage was found to the posterior aspect of the mid cervical area of the spinal cord. There was, however, no evidence of intracranial, optic nerve or retinal pathology and no soft tissue neck injuries to support a traumatic causation.

Nonaccidental head and neck trauma cases [Figure 7]

Head injury case 9 A 3-month-old female child was found dead at home in her bed. The father had allegedly placed her on the bed, unsupervised, with a bottle of milk propped in the child's mouth. At *post-mortem* she was found to have been the victim of previous episodes of physical abuse with a recent head injury (subdural haematomas, optic nerve and retinal haemorrhages). Subdural haemorrhage was present to the entire spinal cord. There were recent injuries to the spinal dorsal nerve roots with associated haemorrhage. There was no epidural haematoma.

Head injury case 10 A 2-year-old female child allegedly collapsed whilst getting changed. She was alleged to have fallen 81 cm into a ditch earlier in the day but had been clinically well following the alleged incident. At *post-mortem* she was found to have both old and fresh external

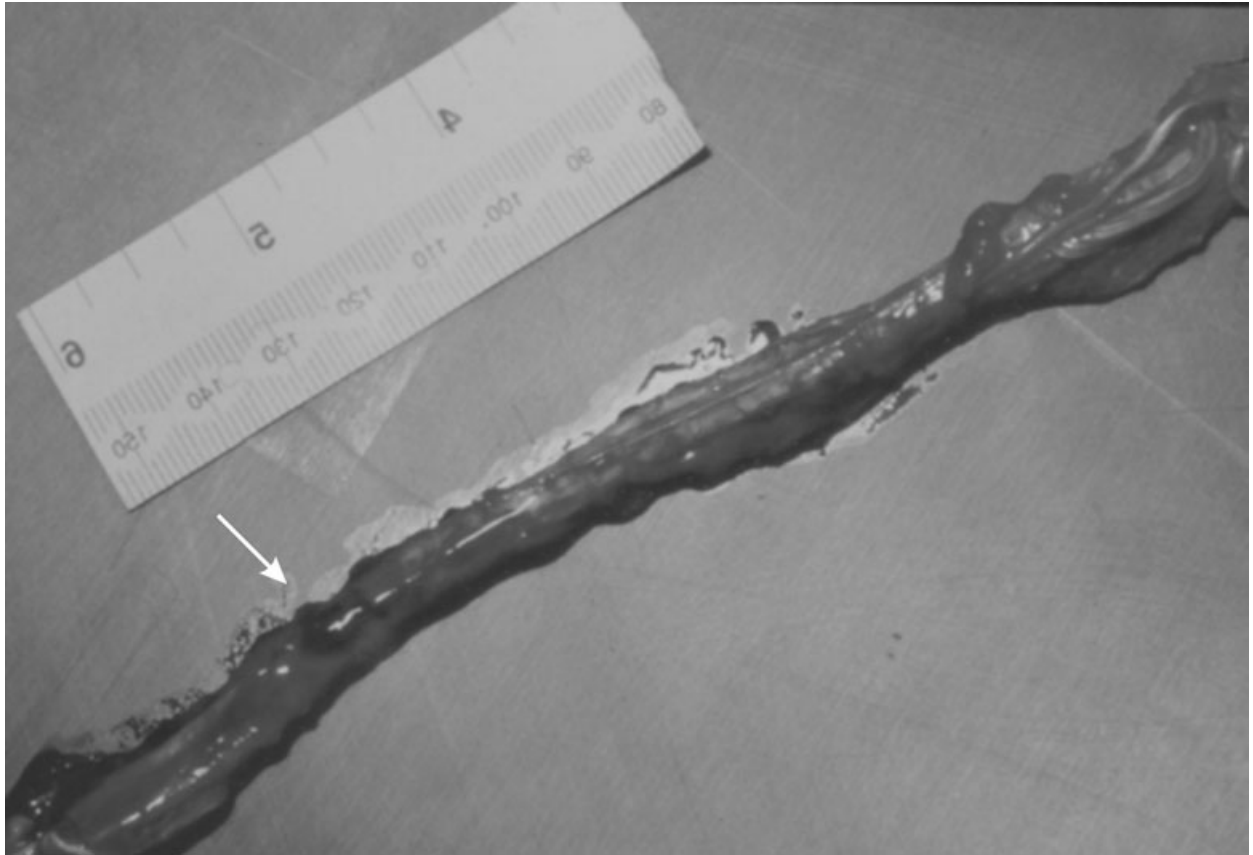


Figure 6. Localized area of posterior epidural cervical haemorrhage (arrow) in a case with a history of physical abuse but no head or neck trauma at death.

bruising with an old internal abdominal injury. She had both old and fresh subdural haematomas, a deep soft tissue facial injury, optic nerve and retinal haemorrhages. Bruising was present to the soft tissues of the posterior aspect of the neck at the level of C3. Subdural haematoma was present to the entire spinal cord. Cervical nerve root damage was present. There was no evidence of epidural bleeding.

None of the cases presented had an abnormality of their coagulation, haematology, immunology, microbiology, virology, metabolic, chromosomal or toxicology studies undertaken in life or after death. Where epidural haemorrhage was present it showed no associated *antemortem* nerve root injury, β amyloid precursor protein or haemosiderin staining and was separate from the site of the CSF puncture. All cases had a full skeletal X-ray undertaken prior to *post-mortem* but none were examined using computerized tomography (CT) or magnetic resonance imaging (MRI). However, because of the size of the epidural blood collection it is doubtful (but not proven or

excluded) as to whether any of these collections would have been visualized using these axial imaging techniques.

Discussion

Spinal epidural haematoma was first reported in 1869 and is an infrequent event that usually occurs in adults with a peak age of onset between 60 and 70 years [19]. It is a relatively rare pathology of childhood with most reported cases occurring between the ages of 5 and 14 years with only two cases under the age of 2 years [7–37]. In childhood it has a classic clinical presentation of sudden onset of back pain followed by acute neurological deterioration attributed to cord compression from the accumulation of blood in the posterior or posterior-lateral epidural space. The diagnosis is made by MRI and treatment may require surgical decompression. Although all levels of the spinal column may be affected, most cases occur within the lower cervical, upper thoracic region, usually between T3 and T6.



Figure 7. Postfixation cross sections of the cervical cord showing subdural haemorrhage in a case of nonaccidental head injury.

Many pathologists may approach the neck in both children and adults from an anterior approach although a posterior approach has, at times, distinct advantages. Although the anterior approach allows for a layered anterior compartment dissection, access to the cervical spinal cord, especially the upper segments may prove technically demanding in the absence of a traditional 'y' neck incision; a shoulder-to-shoulder chest incision has been preferred more often in children. We have opted for a single midline posterior incision, starting just posterior to the vertex of the scalp in the midline and extending to the natal cleft, to examine both the cranial cavity and spinal cord. An incision such as this allows for the examination of the scalp, cranial bones, cranial contents, muscles of the back, the lateral and posterior aspects of the ribs as well as the spinal cord. It allows for a layered posterior neck compartment examination in trauma cases and identification and location of any haemor-

rhage to the spinal cord as presented in this paper, which may be missed by the traditional anterior approach. In cases of congenital abnormality, vascular disease, posterior fossa pathology, raised intracranial pressure or consideration of cranio-cervical junction pathology a window can be removed from the occipital bone to observe and allow removal of the cerebellum, brainstem and cervical cord in continuity. Reconstruction following the *post-mortem* is cosmetically more acceptable compared to some other incisions, simple to perform and allows viewing.

The majority of cases of spinal epidural haematoma reported in childhood have no underlying causative factor and have been labelled as 'spontaneous' or idiopathic. The other reported causes are coagulopathies including haemophilia A and B, factor IX deficiency and leukaemia, therapeutic procedures in the form of the use of epidural catheters or spinal taps, and vascular abnormalities such

as arterio-venous malformations or epidural angioma. Towbin, described spinal extradural haemorrhage (EDH), most dense in the cervical region, in seven of eight cases of 'sudden infant death syndrome' [39]. In a later review of 600 neonatal deaths and stillbirths he described EDH as common in this population and assumed to be attributed to trauma, epidural haemorrhage and spinal injury occurring in 10% of babies [40]. Trauma is a rare cause with cases reported in association with fractures of the spinal column and road traffic accidents. Three cases of epidural haemorrhage were reported by Geddes *et al.* in their paper concerning inflicted head injury in children although the description given within the paper is vague with no mention of the location, size or association with the cranio-cervical or nerve root injuries [4]. Hadley *et al.* also reported the presence of extradural haemorrhage to the cervical spinal cord in five of six cases presented as whiplash-shaken injury syndrome although again the descriptions given within the text lack the site, size or association with cranial nerve injuries [41].

To date the pathogenesis of epidural haemorrhage remains unknown. Some have suggested that the bleeding is venous in origin, been derived from thin walled epidural venous plexus, which may be vulnerable to leakage from altered intrinsic pressure. Others have suggested an arterial origin and finally suggestions have been proffered that the bleeding originates from mechanical disruption associated with torn nerve roots or spontaneous rupture of occult arteriovenous malformations.

Although clinically epidural haemorrhage may be life-threatening attributed to cord compression, the presence of the epidural cervical blood presented within this paper does not fall into any of the above clinical presentations. There were no clinical symptoms, no history of *antemortem* head and/or neck trauma (in the non-head and neck trauma cases) and no evidence of coagulopathies. The haemorrhage was located in a posterior or posterior-lateral position but did not have the same shape or volume seen in clinical epidural haematomas and there was no evidence of cord compression. In all cases the haemorrhage appeared fresh. Finally the ages of the children were outside that normally associated with clinical haemorrhage at this site.

So what is the cause of this observation and does it matter? In recent times more emphasis has been placed upon the examination of the spinal cord, specifically the cranio-cervical junction in the investigation of childhood nonaccidental head injury. Geddes herself, as stated above,

draws attention to the finding of epidural bleeding to the cervical spinal cord although, as stated above, because of the lack of description within the paper as to the site of this haemorrhage, no further interpretation can be drawn as to the possible cause of the bleeding or the relationship, if any, to the spinal cord or head injury. As it has been proposed that nerve root tears may be a cause of this haemorrhage it is critical that a thorough examination of the spinal cord is undertaken in all cases of child death. We have observed that an important pathology when considering whether a nonaccidental injury has occurred in the form of a 'shaking/shaking-impact' is the presence of *antemortem* cervical nerve root injuries, caused by hyper-extension/flexion of the neck beyond that of the normal neck movement of the child. This supports previous observations in animals and humans in relation to excessive flexion/extension movements of the neck [42,43]. However, in the two cases presented that had *antemortem* nerve root changes (cases 9 and 10), which occurred in the presence of associated head injuries, epidural haemorrhage of the cervical cord was in fact not found. The bleeding to the spinal cord in such cases occurs in the subdural and subarachnoid space and in our experience is often distributed to the entire length of the cord.

The over-interpretation of epidural haemorrhage in the cervical region may lead to the mistaken diagnosis of nonaccidental head injury. This is illustrated in cases 7 and 8 where the children showed evidence of *antemortem* physical abuse and both had cervical epidural haemorrhage at *post-mortem* although death was not attributed to a head injury for which there was no *antemortem* or *post-mortem* evidence. As illustrated in case 5, even haemorrhage around the cranio-cervical junction must be interpreted with extreme caution as this may occur in the absence of a history or *post-mortem* finding of head and/or neck injury.

The documentation of the site and timing of the taking of a CSF sample is critical so as not to interpret the finding of haemorrhage at the cranio-cervical junction as attributed to nonaccidental injury when in fact it is attributed to the needle puncture procedure carried out in the *post-mortem* period. This bleeding, as is illustrated in Figure 2, can be distinguished from other areas of epidural haemorrhage and may account for the observation of localized subdural haemorrhage to the cervical cord in case 6 where again there was no evidence of *antemortem* or *post-mortem* head/neck trauma. Finally, the possibility that it is

caused by the opening of the spinal column is also rejected as this would be anticipated to produce more diffuse bleeding within the epidural space along the entire spinal cord rather than localized haemorrhage, which would then be left in the space following the removal of the cord, rather than adhering to the dural membrane.

Thus one possibility is that the epidural haemorrhage illustrated in this paper is a *post-mortem* artefact similar to that described elsewhere in the neck. In 1951 Prinsloo and Gordon described bleeding that occurs to the neck principally at the level of the larynx and thyroid cartilage in the *post-mortem* period [38]. The location of the blood is dependent upon the position of the body but in a prone cadaver, most is seen to the retropharyngeal connective tissues. Thus could this be a similar phenomenon to the cervical spine? If one considers the relative position of the occurrence of the bleeding in the epidural space one sees that, in the majority of cases, it approximates to the same level in the neck as the Prinsloo–Gordon artefact although bleeding may occur outside this level as epidural haemorrhage may be seen to other areas of the spine including the thoracic spine (case 2) and the lumbar area (case 5). The blood is posterior or posterior-lateral in location (as is usual clinically), occurring most commonly at a point approximately halfway along an unsupported neck.

We are thus of the opinion that, following the exclusion that this is a true *antemortem* haemorrhage and exclusion of the known causes of epidural haemorrhage that this is a *post-mortem* artefact possibly similar to that of the Prinsloo–Gordon artefact. One simple explanation for its cause is that it occurs in the *post-mortem* period during the discovery and subsequent handling/movement of the child. If the child is dead then it is unlikely that it will be picked up in the same manner as that of a living child where the head and neck would be expected to be supported by the carer. Even if this was the case, the loss of muscle tone produces the 'floppy' child often described by the discovering carer, which could lead to abnormal body movement during handling. Thus it may be that the handling of the dead child may be performed with no or limited neck support thus resulting in abnormal hyper-extension/flexion or lateral movement of the neck with tearing of the thin venous plexus of vessels around the nerve roots (but not the nerve roots themselves), which in turn leads to the gravitational accumulation of blood within the epidural space. Case 1 supports this mechanism as in this case the father was observed not only to be

holding the dead child with its head unsupported but also to possibly 'shake' it in an attempt to resuscitate it. Of all the nontrauma cases illustrated this case had the most pronounced cervical epidural haemorrhage, which may have been caused by these actions in the *post-mortem* period.

Finally, we believe that a possible explanation for the lack of extradural haemorrhage in cases with clearly demonstrable head injury with brain swelling and subdural/subarachnoid haemorrhage may be the increase in pressure within the spinal dural sheath itself thereby tamponading the extra-dural venous plexuses thus preventing extra-dural haemorrhage formation from *post-mortem* hypostasis.

In summary, we present a series of cases to illustrate the finding of epidural haemorrhage principally to the cervical cord discovered at *post-mortem*. This finding must be interpreted with caution. The over-interpretation of this finding as supportive of excessive neck movement as is hypothesized to occur in 'shaken baby syndrome' may lead to a mistaken diagnosis of a nonaccidental injury especially in the absence of an *antemortem* history or *post-mortem* finding of a head injury. The interpretation of the finding related to the clinical history, *antemortem* and *post-mortem* findings and investigations are critical. We have shown that this finding may occur in the *post-mortem* period as an artefact possibly caused by the handling of the child after death and that it may not occur in those cases with demonstrable head injury attributed to the tamponading effect of raised dural sheath pressure on the venous plexuses.

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