Lethal complication in Pott’s puffy tumor: a case report

Hideki Hyodoh¹
Kotaro Matoba¹²
Manabu Murakami³
Atsuko Saito¹
Tomoko Matoba¹

¹ Center for Cause of Death Investigation, Hokkaido University Faculty of Medicine
² Department of Forensic Medicine, Hokkaido University School of Medicine
³ International Relations Office, Hokkaido University Graduate School of Medicine
Abstract

A man in his fifties was found dead in his bed. Using postmortem CT, the frontal sinus wall was seen to have been destroyed and a subcutaneous / intra-cranial mass-like lesion was detected. Postmortem blood biochemical examination demonstrated high values of urea nitrogen, c-reactive protein, procalcitonin, and precepsin, which were thought to be due to sepsis. Needle aspiration showed reddish viscous fluid, and the presence of Klebsiella oxytoca was confirmed by culture inspection. Based on these results, Pott’s puffy tumor with intracranial empyema, and dehydration with sepsis in the agonal period was assessed as the cause of death. Using autopsy evaluation, it was possible to come to a concrete conclusion, but a minimally invasive autopsy might be an alternative approach to investigate the cause of death.

Key words
Postmortem CT; Pott’s puffy tumor; cause of death investigation; Forensic Radiology; Frontal osteomyelitis
Introduction
Paranasal sinusitis is a common disease, and using systemic antibiotic agents, the frequency of severe complications has been decreasing in decades [1]. When sinusitis is deteriorating, inflammation may lead to frontal bone sinusitis / osteomyelitis, and a rare complication, called Pott’s puffy tumor [2, 3], has been reported, which is a painful forehead soft tissue tumor. It may extend intracranially, resulting in a sub-periosteal abscess and subdural empyema [4-6]. Without suitable treatment, the subdural empyema may become the cause of death [7]. In this case report, we inspected the deceased body using postmortem CT without forensic autopsy, and evaluated the postmortem blood examination, from which the Pott’s puffy tumor (and its related complications) was thought to be the cause of death.
Case
A man in his fifties lived by himself. His aunt, who lived nearby, visited his home. But she couldn't make contact with him, so that she decided to inspect his room with a police officer, and they found the deceased body lying in a supine position in bed. This was in mid-January (winter season) and the water, gas, and heating supply had been cut, so that the room temperature was 2 degrees Celsius.
In his room, many nonsteroidal anti-inflammatory tablets were found, and many used tissue papers were discarded beside his pillow.

A summation of findings is presented in Table 1.

Table 1. Deceased body appearance

<p>| | |</p>
<table>
<thead>
<tr>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Height</td>
<td>166cm</td>
</tr>
<tr>
<td>Weight</td>
<td>51.1kg</td>
</tr>
<tr>
<td>Rigor mortis</td>
<td>spread to almost the whole body</td>
</tr>
<tr>
<td>Postmortem lividity</td>
<td>slight dark reddish at the rear, and the color disappeared with compression by tweezers</td>
</tr>
<tr>
<td>Cornea</td>
<td>medium turbidity</td>
</tr>
<tr>
<td>Body surface</td>
<td>no injury</td>
</tr>
<tr>
<td>Forehead</td>
<td>soft tissue tumor-like mass was palpable on the right forehead and bony structure was not palpable at the arcus suprerialis</td>
</tr>
<tr>
<td>Eye</td>
<td>open bilaterally, protrusion</td>
</tr>
<tr>
<td>Rectal temperature</td>
<td>about 30 degrees Celsius (measurement at site of discovery)</td>
</tr>
<tr>
<td>Postmortem interval</td>
<td>about 12 – 24 hours estimated</td>
</tr>
</tbody>
</table>

After bringing in the deceased body to our department, the whole body was evaluated by computed tomography (16-slice multi-detector CT scanner (Supria, Hitachi Corp., Tokyo). The scan parameters were as follows: 120 kV, 215 mA, 0.75 s/rotation, beam pitch 1.3125, collimation 1.25 × 16, slice thickness 5.0 mm).

Postmortem CT findings:
A soft tissue mass was found at the right frontal sinus with osteolytic change at the inner and outer bone structure of the frontal sinus, and it showed expansible extension into the cranium of about 5 cm (Figure 1). The osteolytic change was also found at the frontal skull base and reached into the sella turcica. The upper extension was about 5 cm from the frontal skull base, and the bilateral frontal cerebral lobe presented lower density change in deep white matter, estimated as edematous change. The mass margin
was clear and smooth, and a lump was visible at his forehead. The mass extended into both orbits. The osteolytic change was found at the right inner bone structure, and the right inner straight muscle and optic nerve were compressed externally. The left inner bone structure was compressed by the mass lesion, and the left retro optic structures were compressed to the frontolateral side. The mass extended into the ethmoid sinus and right maxillary sinus, so that the mass border was not clearly distinguishable from membranous swelling. The inner density was inhomogeneous and calcified density was found (Fig. 1a, b).

The lung finding on postmortem CT presents no hypostasis in bilateral lung field, and the lung aerated area (minus 1000 to minus 700 HU) was calculated as about 94% (Fig. 2b).

Postmortem blood examination is presented in Table 2.

Table 2. Postmortem blood examination

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Value</th>
<th>Normal Clinical Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carbon monoxide concentration of Hemoglobin</td>
<td>0.4 %</td>
<td>(0.5 - 1.5)(1.5 - 5.0 smoker)</td>
</tr>
<tr>
<td>Hemoglobin A1c</td>
<td>6.5 %</td>
<td>(4.7 - 6.2)</td>
</tr>
<tr>
<td>Blood urea nitrogen (BUN)</td>
<td>304.7 mg / dL</td>
<td>(8 - 20)</td>
</tr>
<tr>
<td>C reactive protein (CRP)</td>
<td>16.95 mg / dL</td>
<td>(≦0.30)</td>
</tr>
<tr>
<td>N terminal pro B-type natriuretic peptide (NTproBNP)</td>
<td>1220 ng / L</td>
<td>(≦ 18.4)</td>
</tr>
<tr>
<td>Procalcitonin (PCT)</td>
<td>2.36 ng / mL</td>
<td>(≦ 0.05)</td>
</tr>
<tr>
<td>Precepsin</td>
<td>2970 pg / mL</td>
<td>(≦ 500)</td>
</tr>
</tbody>
</table>

(): normal clinical value

Postmortem urine toxicological examination
Using triage simple inspection test, all items presented negative.

Lesion aspiration and culture inspection
The right forehead lesion was punctured using an 18-gauge spinal needle, and aspirated a red viscous liquid. No solid component was collected and an abscess was confirmed. The culture inspection confirmed *Klebsiella oxytoca* infection.

According to the postmortem CT findings, the soft tissue mass was found at the right frontal sinus with osteolytic change at the skull bone, and a mass-like lesion was presented in the intracranium. In addition, lung hypostasis was absent and potential differential diagnoses included dehydration, loss of a lot of blood, hypothermia, and unusual death position such as standing (strangulation) [8].
From the postmortem blood and urine examination, the high BUN value was thought to be affected by dehydration, and this was thought to be due to sepsis, because of the high CRP, PCT and Presepsin values. From these results, with further confirmation by aspiration and culture inspection, Pott’s puffy tumor was diagnosed, and intracranial extension via vein and direct osteolytic extension into the subdural space, which may present subdural empyema, and dehydration with sepsis in the agonal period, were suspected.
Discussion

Paranasal sinusitis is recognized as one of the high frequency diseases, and one of the rare complications, involving sinusitis characterized by osteomyelitis of the frontal bone with subperiosteal abscess, was named Pott’s puffy tumor, as it was first described by Sir Percival Pott in 1768 [9]. Since the advent and widespread prescription of antibiotic therapy, Pott’s puffy tumor has become a rare entity [4, 10]. Intracranial complications with or without direct erosion of the frontal bone have been observed in about 60% to 85% of these patients [11-15]. The pus may erode the bony walls of the sinuses, directly invading the leptomeninges, or bacteria may infect the bridging veins that run through the bony calvarium and leptomeninges, carrying bacteria along the veins into the leptomeninges [16]. In addition, venous drainage of frontal sinuses occurs through diploic veins that have communication with the dural venous sinuses, so that the intracranial complications occur either due to direct extension or venous drainage [1].

The several chronic diseases that affect the innate immune response, such as diabetes, chronic renal failure, and anaplastic anemia, or cocaine inhalation could be risk factors for the development of Pott’s puffy tumor [1, 13, 17]. In our case, not only frontal bony wall distraction, but also periosteal, and intracranial abscess were observed by postmortem CT, so that these findings were concordant with the reported cases.

The most frequent bacterial agents involved are *Staphylococcus aureus, Streptococcus spp,* and *anaerobes* [18-20]. In this case, reddish pus was collected from the lesion, and its cultural evaluation confirmed Klebsiella oxytoca infection. In addition, postmortem blood examination showed high values for PCT, CRP, and Precepsin, so that severe sepsis occurred in the agonal period. It is considered highly probable that frontal sinusitis developed to a chronic stage leads to frontal bone osteomyelitis, periosteal abscess formation (Pott’s puffy tumor), and intracranial abscess extension, which is thought to lead to lethal septic shock.

CT is a useful evaluation tool in postmortem status [8]. Because the deceased body was dehydrated, there is no lung edema and no hypostasis (lung aeration was maintained) in this case. When the postmortem CT shows high aeration in lung field, the differential diagnoses might be dehydration, hypothermia, neck hanging (malposition), and loss of blood [8]. In this case, the deceased body’s rectal temperature was 30 degrees Celsius when measured at the site of discovery lying in bed in a supine position, without blood loss, so that dehydration remained among the differential diagnoses. In this case,
postmortem CT could elucidate the body condition in the agonal period, showing that the dehydration occurred due to the intracranial abscess formation and lethal septic shock.

If this lesion was found clinically, contrast enhanced CT might be useful to demonstrate the congestion at the marginal region of the abscess and detect another complication [21]. Use of postmortem CT angiography to evaluate the vascular structure has been reported, and it is superior to autopsy and CT without angiography to help detect forensically essential findings [22]. However, postmortem CT has not been used extensively to assess for intracranial inflammation, so that the usefulness of using contrast to detect inflammation (or abscess) in postmortem CT is not yet confirmed. Magnetic resonance imaging could demonstrate inflammation (or abscess) without contrast [1, 23], so that it might be useful in evaluating a deceased's condition.

There still remain unsolved questions, like whether the lesion might contain solid tumor which occludes the nasal canal, and lead to para-nasal sinusitis, and whether a brain abscess might occur due to the effect of dural inflammation. It is thought that a full autopsy evaluation would be necessary in order to confirm the tentative evaluations and lead to a more reliable concrete conclusion.

Using postmortem CT combined with sample analysis (such as the biochemical and toxicological examination of urine and blood), we could diagnose Pott’s puffy tumor as the cause of death, and postmortem CT with multidisciplinary examination could evaluate the deceased terminal condition objectively. A minimally invasive autopsy might be an indispensable approach to cause of death investigation.
Figure 1
Postmortem CT, multi-planar reconstruction, (a) soft tissue window (WL 40, WW 300), (b) bone window (WL 450, WW 1500), and (c) 3-dimensional reconstruction (volume rendering).
Soft tissue mass-like lesion presents at the frontal sinus as epicenter (a), and osteolytic change is demonstrated at the frontal sinus wall, frontal skull base, and Sphenoid sinus (b). The lesion also extends into the orbital region, and compresses the structure, laterally. The bilateral eyes are displaced due to the tumor mass effect. Osteolytic change is detected at the right arcus suprachiliaris.

Figure 2
lung
There is no hypostasis in bilateral lung field even though the deceased was found in a lying position. The lung aeration remained and no pleural space fluid was detected. Pneumomediastinum was also detected.
Figure 1-a. Postmortem CT, soft tissue window (WL 40, WW 300)
Figure 1·b. Postmortem CT, bone window (WL 450, WW 1500)
Figure 1c. 3-dimensional reconstruction
Figure 2. Postmortem CT, lung
References

[21] Paranasal lesion Diagnostic Imaging Guideline, 2007. [http://www.radiology.jp/content/files/406.pdf#search=%27%E5%89%AF%E9%BC%BB%E8%85%94%E7%82%8E+%E7%94%BB%E5%83%8F+%E3%82%AC%E3%83%A4%E3%83%89%E3%83%A9%E3%82%A4%E3%83%B3+%E8%A%9E%27].