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Case Report

Pathology Section

Lipoid Pneumonia and Peritoneal Endometriosis: Rare Incidental Findings on Autopsy

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ABSTRACT

Lipoid pneumonia has been described in as early as 1925, by Laughlen GF, to be a rare variant of pneumonia caused by the inhalation of oil droplets. Around 50% of the patients with lipoid pneumonia are asymptomatic. Here, a case is presented where peritoneal fluid and tissue pieces from various organs of a 12-year-old female were received for examination after autopsy. There was no history of any illness before death. Cytology of fluid revealed features suggestive of endometriosis. Microscopy of lung sections revealed numerous lipid droplets in the alveoli and the interstitium along with a mild inflammatory infiltrate suggestive of lipoid pneumonia. In view of incidental findings in present case report, the occupational history or history of inhalation of oil-based medications becomes very important and should always be investigated. Also, in the absence of any symptoms peritoneal fluid needs to be studied carefully for any pathology.

Keywords: Alveolar macrophages, Lipid droplets, Peritoneal cytology

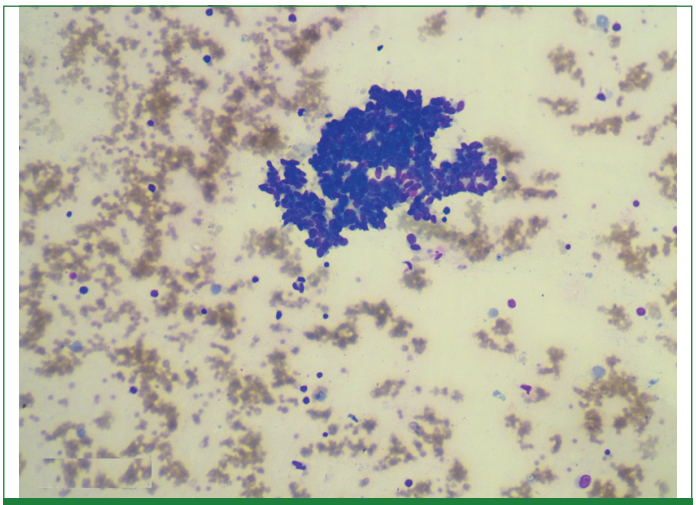
CASE REPORT

A 12-year-old female, who was unwell for five to six days with the complaint of fever and was being treated at a private clinic. After her condition worsened, she was brought to the tertiary care centre and was managed conservatively, where she succumbed on the same day. Autopsy was performed, as it was a medico-legal hospital death.

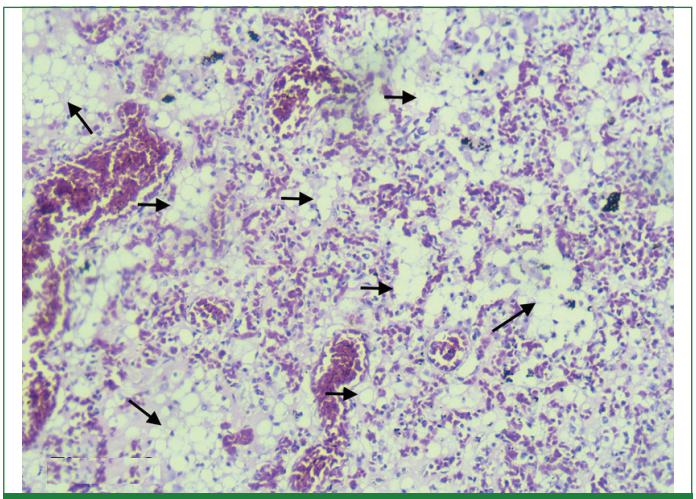
Subdural haemorrhage was noted and the brain parenchyma was unremarkable. Both lungs were congested and blood tinged froth oozed out on cut section. Pericardial cavity and pericardium were unremarkable. Heart, coronary arteries, great vessels and valves were unremarkable. All the coronary ostia were patent. Peritoneal cavity had about 300 cc of reddish brownish coloured fluid which was sent for cytological examination. Liver weighed 1231 gm and was grossly unremarkable. Spleen was enlarged and weighed 174 gm and was congested. Pancrea was unremarkable. Stomach had 5 cc of yellowish content. Small intestine contained fluids and gases. Large intestine contained faecal matter and gases. Mucosa of both small and large intestine was unremarkable. Pelvic cavity was unremarkable. Urinary bladder and the rectum were empty. The uterus was menstruating. Both sides adnexal organs were unremarkable.

Smears were made from the peritoneal fluid that was sent for cytological examination and stained with Geimsa stain. The smears were highly cellular with two population of cells. One type of cells was reactive mesothelial cells. These were present in loosely cohesive sheets showing mild atypia with few having nuclear grooves. The second type of cells was identified as endometrial cells. These cuboidal cells were present in small clusters along with occasional small sized spindle cells [Table/Fig-1]. The background was haemorrhagic. On the basis of these findings and supportive postmortem findings of blood tinged haemorrhagic peritoneal fluid and menstruating uterus, a diagnosis of peritoneal endometriosis was suggested.

Multiple sections stained with Haematoxylin and Eosin (H&E) of brain, liver, kidney, spleen, heart and lungs were also examined. Sections from brain, heart, kidney, spleen and liver revealed no significant pathology. Multiple sections from lungs showed markedly congested blood vessels. Numerous intra-alveolar macrophages containing distinct intracytoplasmic vacuoles of varying sizes resembling lipid droplets were also noticed [Table/Fig-2]. These lipid droplets were also seen in the interstitium. Mild mononuclear inflammatory cell



[Table/Fig-1]: Cluster of endometrial cells with few scattered stromal cells in a haemorrhagic background (Geimsa stain, 200X).



[Table/Fig-2]: Sections from lungs showing markedly congested blood vessels, numerous intra-alveolar macrophages containing distinct intracytoplasmic vacuoles of varying sizes resembling lipid droplets (H&E, 100X).

infiltrate in the interstitium was also seen. Occasional plasma cells were also present. However, no fibrosis was found. Based on the histopathological features, possibilty of lipoid pneumonitis was suggested. Since, these were the findings of an autopsy case, no further investigation or confirmatory tests could be conducted for both of the diagnosis.

DISCUSSION

'Autopsy' word is derived from ancient Greek word 'autopsia' which means to see for oneself [1]. Hence, the histopathological examination of autopsy specimens contibute greatly to the vast knowledge of existing diseases and also to the pool of rare conditions. Histopathological examination always presents with interesting and rare conditions for pathologists to study and learn. It, thus, becomes a dynamic and continous source of learning and evolving. Lipoid pneumonia is one such rare variant of pneumonia. It was first described by Laughlen GF in the twentieth century in one adult, one infant and two children. He reported lipoid pneumonia to occur after repeated inhalation of nasopharyngeal oil droplets [2].

In 1984, Becton DL et al., reported an interesting case of lipoid pneumonia in a young girl due to use of lip gloss, earlier all case reports had been of older individual above the age of 50 years [3]. In autopsy studies, Baron SE et al., have given its incidence to be 1.0-2.5% [4]. Lipoid Pneumonia (LP), is described as an uncommon disease caused by deposition of lipid in alveoli and interstitium [5]. It is classified as exogenous, endogenous and idiopathic based on the mode of lipid deposition. The exogenous type is the most common and the idiopathic variant is the rarest, which is also associated with smokers [6].

Various case reports have suggested a varied age group, Becton DL et al., reporting in a 18-year-old girl to Rana D et al., reporting in 53-year-old male [3,7]. The authors report the present case in a 12-year-old girl. Several authors have cited multiple causes for deposition of lipid as: gas inhalation [8], oil based medications for respiratory diseases [9], aspiration of milk [10], poppy seed oil [11], egg yolk [12], use of spray paint [13], longstanding use of petroleum jelly (vaseline, vicks) at bedtime [14], excessive use of lip balm (chap Stick, a lipstick that contains petrolatums and lipids) and of flavoured lip gloss [3]. The clinical symptoms greatly vary according to the type of oil and duration of exposure. The symptoms are non specific and present as chronic pneumonitis with chronic cough and progressive dysponea. Lipoid pneumonitis may be completely asymptomatic or can present as mild disease [15]. Often the diagnosis is based on history of any exposure to oil products, but it is either missed or difficult to establish [16]. Radiological findings in LP are too non specific. The most frequent findings are ground-glass opacity or consolidation of the lower lobes as seen in infective community-acquired pneumonia. Fibrosis and coalescence of oil droplets can result in nodules and masses with irregular margins, closely mimicking lung cancer [16]. In such cases Bronchalveolar Lavage (BAL) and biopsy clinches the diagnosis. However, no such foci of carcinoma were seen in the present case. The BAL reveals lipid laden macrophages, however presence of extracellular oily droplets is more specific for LP. Histopathological features suggestive of LP are presence of lipid-laden macrophages that fill and distend the alveoli and interstitium, accumulation of lipid material in interstitium, inflammatory infiltration as observed in the present case [17].

Fibrosis is another most significant finding seen in many cases [18]. However, neither fibrosis, nor any foci of carcinoma were observed in the present case. The diagnosis can be confirmed on frozen sections by using stains specific for lipids, where vacuoles are stained in orange with Sudan III, brownish-orange with Sudan IV, and deep red with Oil Red O. Sudan black B stains cholesterol esters and triglycerides in dark blue, and some phospholipids in gray [18]. In the present case, any special stain could not be attempted as all the sections were formalin fixed and paraffine embedded. Also, in the present case the patient did not have symptoms of pneumonia and thus, lipoid pneumonia was detected on autopsy. The pathophysiology of lipoid pneumonia is not completely understood. Graef I has described in his article that oil droplets do not initiate a cough reflex because of bland nature of oil. When these oil droplets reach the lower airways, they inhibit the mucociliary clearance and are deposited in the alveoli and interstitium. Humans lack the enzymes to metabolise lipid hence these lipid droplets are engulfed by macrophages and stay there, slowly releasing lipid in interstitium upon disintegration. Following this a giant cell reaction is initiated and on long standing fibrosis of tissue occurs [18]. Fibrosis was probably not seen in the present case as the patient was young and may be the duration of exposure to the inciting agent was not long enough.

Another incidental finding in the present autopsy was that, the authors reported was peritoneal endometriosis. Endometriosis is common entity with ovarian endometriosis (46%) being the most common followed closely by peritoneal endometriosis [19,20]. Other uncommon sites include extrapelvic locations like cervix, vagina, vulva, intestinal tract, urinary tract, abdominal wall, thoracic cage and lungs [21]. Markham SM et al., reported endometriosis in their study to be present in every tissue except spleen [22]. It primarily affects women in the reproductive age group. The most frequent symptoms include dysmennorrhoea, chronic pelvic pain and infertility. The patient being very young must have yet not experienced any of the symptoms. The utility of peritoneal wash cytology for diagnosis of endometriosis has been reported widely [23,24]. In most cases, only haemosiderin-laden macrophages are identified though the presence of endometrial cells is more specific [24-27]. The endometrial cells have been reported in around 25-52% of peritoneal wash samples examined [23,27]. Peritoneal fluid contains mesothelial cells that need to be differentiated from endometrial cells. The distinction of endometrial cells from mesothelial cells as given by Stowells SB et al., helps in identifying both types and reach a conclusion [25]. Stowell SB et al., had tabulated the differences between mesothelial cells and endometrial cells which helped us to differentiate between the two population of cells that we saw on the smears from our case [25]. Confirmatory diagnosis for endometriosis is generally made on laparoscopy, but since the present case was an autopsy case, no records could be traced.

Both the incidental findings on the autopsy were probably asymptomatic because the patient was very young to have manifested the symptoms.

CONCLUSION(S)

The rarity and non specific findings of Lipoid pneumonia makes it a condition that can be misdiagnosed clinically. It leads to pulmonary fibrosis and hence, can be a cause of significant morbidity to patients, thereby making its early recognition and diagnosis important. The clinicians need to be aware of this entity while assesing the radiological findings of delayed, persisting pneumonia and also take note of important exposure history. Early diagnosis will lead to targeted therapy and save the patient from unnecessary antiobiotic exposure, whereas peritoneal endometriosis is a much commoner entity. The peritoneal wash specimens in females with supporting history must be examined thoroughly. The attentive examination of specimen in the present case report led to diagnosis of lipoid pneumonia, a rare entity and peritoneal endometriosis, a common entity that presents a good learning exercise during evaluating autopsy specimens.

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