

Exceptional dissemination of perineal tuberculosis up to the right flank: a tribute to J.P. Nesselrod's study on the anatomy of pelvic lymphatics published in 1936 in the *Annals of Surgery*

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Case

A 56-year-old cachectic male in poor general and hygienic condition collapsed and was subsequently admitted to hospital in a severe sepsis. The personal history was negative for preexisting intraabdominal and cardiorespiratory ailments. Recent journeys to tropical countries were denied. However, a lengthy stay at a psychiatric clinic due to alcohol abuse several years ago was noteworthy. The patient had been suffering from perineal fistula for years without any medical treatment. At admission, we found a complex of

perianal and gluteal abscesses with involvement of the skin of the right lower back. The abdomen was clinically unsuspecting. The laboratory studies revealed a severe inflammatory syndrome measuring a white cell count of $30 \times 10^9/L$, CRP value of 226 mg/L, and slightly elevated procalcitonin with 0.47 $\mu g/L$.

A CT scan revealed an extensive subcutaneous abscess on the right flank with involvement of the right iliopsoas muscle (Fig. 1), the presacral space, the fossa ischiorectalis with involvement of the external sphincter, and the levator ani (Fig. 2a–c) with subcutaneous fistula in the perianal region and the adjacent right gluteus. However, no internal orifice was found by rigid rectoscopy. An endoanal ultrasound to investigate the presence of trans- or intersphincteric fistula was omitted due to the patient's bad condition. The MRI performed to analyze whether there was an involvement of vertebral bodies or an involvement of intervertebral disks was not suitable to evaluate the anal sphincter.

There were no particular intraabdominal findings such as significant lymphatic nodes, wall thickening of the bowel, or ascites. All findings were restricted to the extraperitoneal space.

Despite immediate surgical drainage of the subcutaneous abscess on the right flank and in the perianal region as well as antibiotic treatment with piperacilline/tazobactam, the patient's condition showed signs of deterioration such as weight loss, hypotension, and fever. Additionally, upcoming perianal abscesses were surgically evacuated, and the presacral space was partially drained initially by CT-guided puncture and finally emptied by open surgery over an extraperitoneal right–anterior access to the right iliopsoas muscle. The drained perianal wounds did not show any signs of healing and continued to produce serous turbid secretion. Crohn's disease was suspected early, but not confirmed in an ileocolonoscopy. The microbial culture from

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Fig. 1 The patients' CT scan at admission, showing a large subcutaneous abscess (*star*) on the flank at the level of the fourth lumbar vertebral body, communicating with the iliopsoas muscle, the paraspinous muscles, and the epidural space (*arrow*)

the wound secretion demonstrated growth of *Streptococcus intermedius*, whereas the surgically obtained specimens from the right presacral abscess evolved to be sterile. Serological screening for syphilis and HIV was negative. In a histological sample from the perineal region, tuberculoid granuloma was found, while an interferon gamma release assay (Quantiferon Gold) in the blood was positive. The patient did not suffer from any pulmonary symptoms, but a CT scan of the chest showed reticulonodular infiltrates typically suggestive of pulmonary tuberculosis. Microscopy of the sputum was negative for acid-fast bacilli, but the culture of surgical probes from the perineum and the retroperitoneal space finally became positive for *Mycobacterium tuberculosis* about 1 month after admission. The patient was prescribed a classic tuberculostatic treatment combining rifampin, isoniazid, pyrazinamide, and ethambutol. We observed a rapid healing of the wounds, a significant increase of the patient's body weight, and recovery of the general condition.

Discussion

It is estimated that abdominal tuberculosis accounts for about 1 % of all extrapulmonary manifestations of tuberculosis (TB) [1, 2]. However, the true rate of abdominal involvement might be underestimated [3, 4]. Perianal tuberculosis is reported to be rare and mainly referred to as chronic fistula formation or ulceration related to pulmonary TB [1, 5]. In one series of anal sepsis, TB was found in about 1 % of all cases [5]. In everyday life, the general surgeon is often confronted with perineal abscesses caused by common bacteria of the intestinal tract. In more developed countries, complex fistula formation is mainly suggestive of Crohn's disease that mimics TB in the abdominal cavity as well as in the perianal region [3]. Abdominal TB most often appears with abdominal pain and distension, weight loss, anorexia, and fever. Intestinal bleeding due to ulceration, intestinal perforation, or obstruction are possible complications that urge a surgical intervention. The ileum, the ileocecal region (including mesenteric lymph nodes), and the peritoneum are found to be involved most frequently [6, 7]. Computed tomography findings correlate with the clinical findings: wall thickening of the small bowel, mesenteric lymphadenopathy, and ascites are usually reported from patients with abdominal TB [3, 8].

In our case, an evident involvement of abdominal organs was absent. The patient never had abdominal discomfort, the ileocolonoscopy was normal, and in the CT scan, signs of intestinal or peritoneal involvement were absent.

Anatomically, the extension of the disease was restricted to the right iliopsoas muscle and surrounding presacral tissues, as well as to the perianal region. Although we repeatedly examined the patient clinically, we were not able to find any fistula passing the anal sphincter. However, we

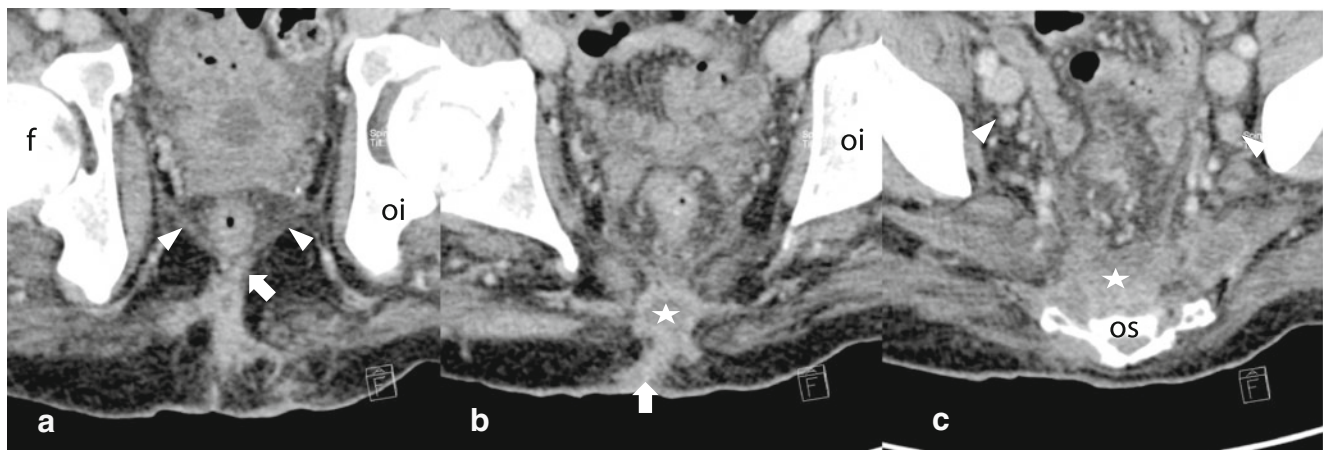


Fig. 2 Coronal reconstruction of the patients' CT scan on the level of the femur (*f*) and os ischium (*oi*). Panel **a** shows a subcutaneous fistula (*arrow*) reaching the levator ani (*triangles*). In **b**, the subcutaneous

abscess (*star*) is linked over a fistula (*arrow*) with the skin. In **c**, the presacral space is filled with abscess (*star*) whereas bilateral lymph nodes are visible (*triangles*)

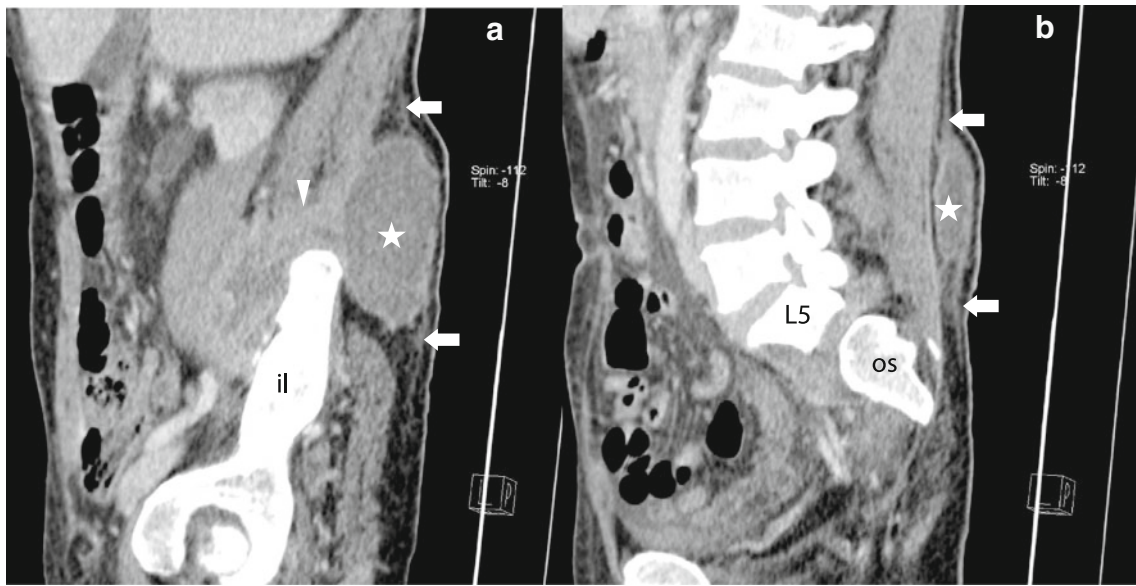


Fig. 3 Sagittal reconstructions of the patient's CT scan at admission. On **a**, the subcutaneous abscess (*star*) and its breakthrough to the iliopsoas muscle (*triangle*) over the right crista iliaca (*il os ilium*) are

shown. A thin subcutaneous spur (*arrows*) delineates potential routes of disease expansion (**b**) on the level of the lumbar vertebral column (*L5* fifth lumbar vertebral body, *OS* os sacrum)

were astonished at the bizarre subcutaneous abscess formation on the right flank that was the leading finding at admission to our hospital. An initially suspected spondylodiscitis could not be confirmed by repeated MRI scans. Overall, we interpret the presented case of perianal tuberculosis as a hematogenous dissemination of pulmonary tuberculosis.

Numerous studies have evaluated the practical consequences of perianal lymphatic drainage in the context of malignant disease. It is well established that tumors below the linea dentata drain to the inguinal lymph nodes [9–11]. Our patient showed no significant inguinal lymphadenopathy, neither in the clinical exam nor in the repeated CT scan.

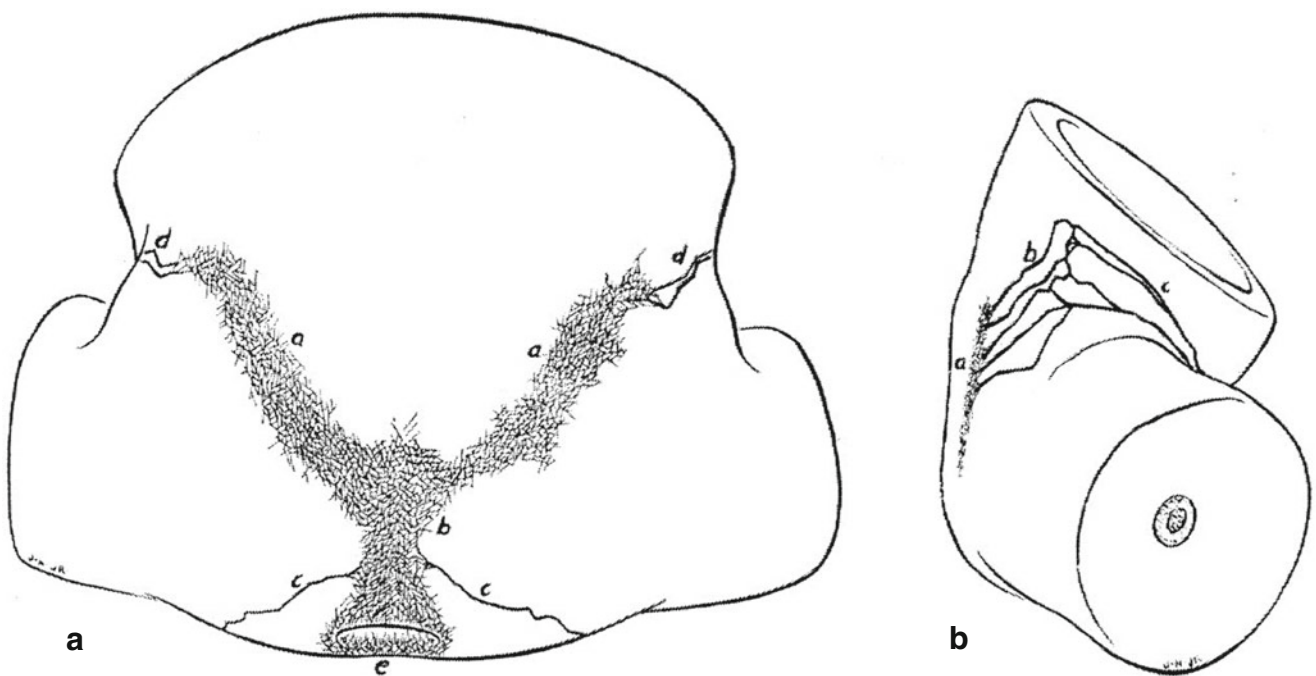


Fig. 4 Original figure from Nesselrod's publication in 1936 [12] in *Annals of Surgery* showing the perineal and gluteal distribution of lymphatic vessels after injections of mercury in the skin, respectively. Panel **a** shows a dorsal view of the sacrococcygeal and superior gluteal region of a

male white fetus at term (original description). Perianal (*e*), presacral (*b*), gluteal plexuses (*a*) and anastomotic vessels (*d*). Panel **b** shows a lateral view on the same specimen as in **a**, showing not only lymphatic vessels above the crista iliaca but also their link (*c*) to the inguinal zone

But the iliacal axis was certainly involved (Figs. 2c and 3a). While searching for an explanation for the exceptional extension and localisation of the inflammatory process involving the subcutaneous tissue on the right flank and the right iliopsoas muscle, we came across an anatomic study published by J.P. Nesselrod in 1936 ([12]; Fig. 4).

The eminent proctologist revealed the existence of anastomotic lymphatic vessels between the perianal skin and a specific gluteal plexus over the buttocks and further backwards to the flank above the crista iliaca. Nesselrod conducted studies with cadavers of human fetuses at or near term, of newborn infants, as well as few but apparently not useful studies on dogs and humans in vivo. He used different dyes, whereby the best and depicted results (Fig. 4) were achieved with mercury which had been injected in the perianal skin. As shown in Fig. 4a, there is a perianal network linked with another network over the crista iliaca over gluteal and sacrococcygeal plexuses. Over the supra-iliacal plexuses, there is a continuing connection to the inguinal nodes hereby encircling the limb. Nesselrod also describes some minor anastomotic lymphatic vessels that pass the anorectal line and ascend in the columns of Morgagni in the ampulla recti. Unfortunately, he does not state whether the iliacal axis was involved upon injection in the perianal plexus.

It is astonishing per se that such an abscess lies subcutaneously on the right flank. This is rare and requires an explanation. Since it was a case of secondary tuberculosis, it must have been a consequence of hematogenous dissemination and not, as it was possible in the case of true intestinal tuberculosis, as a primary infection of the intestine. Principally, it could have been a direct seeding into the right flank. It has to be underscored that the most frequent extrapulmonary manifestation of tuberculosis is the lymphatic form [13]. Therefore, lymphatic networks represent an anatomic space of preferential “homing” for tuberculosis. The sheer presence of lymphatic vessels or plexuses above the crista iliaca made the manner of TB seeding and survival at that precise location more plausible.

Of course, the direction of disease dissemination (break-through through the abdominal flank into or out from the iliopsoas muscle) cannot be definitively cleared. Nesselrod himself reports that perianally injected dyes rise up to the buttocks, but can also descend from the buttocks to the perianal region. Consequently, it is a bidirectional system. Because of anamnestic hints, one has to assume that the primary focus was a perianal abscess that probably took both routes. In our case, there were minor subcutaneous fistulas forming spurs up to the right gluteus (Fig. 3a and b). Internally, CT scan provides clear evidence of the involvement of the puborectalis zone and the iliacal axis (Fig. 2). The iliacal axis only could

have been reached over the involvement of the puborectalis zone. What Nesselrod does not show are anastomotic vessels through the abdominal wall. So it seems reasonable to assume that there were two ways of dissemination (an external and an internal one) that completed an anatomic circle and broke a true anatomic barrier by passing the fascia scarpa and connecting the plexuses above the crista iliaca and the lymphatics of the iliacal axis.

Correlating these old studies with our anatomic findings, we see the clinical relevance of these lymphatic vessels on the buttocks by their concrete involvement.

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Conflict of interest None.

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