

Cholangiosepsis Caused by Neutrophilic Cholangitis in Plaque Psoriasis

Carina Bergthaler^{1*}, Ludwig Kramer², Andreas Steiner¹, Paul Sator¹

¹Department of Dermatology, KH Hietzing, Vienna, Austria

²Department of Medicine, KH Hietzing, Vienna, Austria

Email: *carinabergthaler@hotmail.com

Received 11 January 2016; accepted 7 May 2016; published 10 May 2016

Copyright © 2016 by authors and Scientific Research Publishing Inc.

This work is licensed under the Creative Commons Attribution International License (CC BY).

<http://creativecommons.org/licenses/by/4.0/>



Open Access

Abstract

Psoriasis is a common inflammatory skin disease with many comorbid conditions. We present a 37-year-old male patient with a history of plaque psoriasis, status febrilis, violent umbilical pain, elevated inflammatory markers and liver parameters. Blood cultures were tested positive for *E. coli*. Diagnostic findings indicated that mechanical icterus and cholangiosepsis in the context of neutrophilic cholangitis were caused by inflammatory stenosis. Neutrophilic cholangitis is often found in combination with skin diseases with intense cutaneous infiltration with polymorphonuclear leucocytes and peripheral blood neutrophilia. Interleukin-8 may play a role in the pathogenesis of neutrophilic cholangitis occurring in patients with psoriasis [1]. We showed that complications of psoriasis can also occur at unusual locations. To date no case of neutrophilic cholangitis as an elicitor of cholangiosepsis has been reported.

Keywords

Plaque Psoriasis, Cholangitis, Cholangiosepsis

1. Introduction

Psoriasis is a common chronic inflammatory skin disease affecting approximately 2% - 3% of persons of European descent [2]. In the past decade many studies focused on comorbid conditions in psoriasis. Recent findings show that psoriasis is a systemic disease. Participation of the joints, cardiovascular system and nervous system, diabetes, hypertension, dyslipidemia and inflammatory bowel disease have been found at a higher prevalence in psoriasis patients compared to the general population (Table 1) [3]. Because of the growing range of comorbid conditions, it is important to think of a possible association between psoriasis and other disorders.

*Corresponding author.

Table 1. Psoriasis-comorbidities as a consequence of systemic inflammation [3].

Psoriasis Comorbidities	Studies (N)	Prevalence (%)	95% CI (%)
Anxiety	11	30.2	21.7 - 38.8
Depression	21	21.7	15.1 - 28.3
Psoriasis Arthritis	34	24.1	19.3 - 29.0
Diabetes	21	8.5	7.4 - 9.6
Hyperlipidemia	7	7.4	6.5 - 8.4
Hypertension	20	21.2	19.2 - 23.3
Inflammatory Bowel Diseases	3	0.8	0.1 - 1.4
Adipositas	9	11.9	7.2 - 16.8
Cardiovascular Diseases	12	10.2	7.7 - 12.8

We add a case of cholangiosepsis caused by neutrophilic cholangitis.

2. Case Report

We present a 37-year-old male patient with a medical history of plaque psoriasis. He had one episode of psoriasis in his youth, followed by a long period without a flare. In the past year, he presented with new skin lesions—to date without therapy.

The patient presented with status febrilis and complained of violent umbilical pain. Laboratory investigations showed: elevated inflammatory markers (CRP 218.9 mg/l, white blood count 12.58 G/l) and raised levels of liver parameters (ASAT 153 U/l, ALAT 195 U/l, γ -GT 464 U/l, bilirubin 7.59 mg/dl, LDH 293 U/l, alkaline phosphatase 393 U/l). Blood cultures were tested positive for *E. coli*.

Ultrasound scan showed considerable steatosis hepatis and a marginal splenomegaly. The gallbladder had normal size, pencil-thin wall without intraluminal stones and inconspicuous bile ducts.

Magnetic resonance cholangiopancreatography (MRCP) detected high-grade short distance stenosis of the principal bile duct without prestenotic dilatation or evidence of concretion (Picture 1).

The performance of an endoscopic retrograde cholangiopancreatography (ERCP) showed the stenosis restituted by forced administration of a contrast agent. These findings show an inflammatory stricture and no stenosis caused by a tumor (Picture 2).

The patient's general condition quickly improved and lab parameters returned to normal levels after he received antibiotics (Ciprofloxacin i.v.). Due to the clinical development, we prescinded from invasive examinations.

Diagnostic findings indicate that mechanical icterus and cholangiosepsis in the context of neutrophilic cholangitis were caused by inflammatory stenosis and that complications of psoriasis can also occur at unusual locations.

At the follow-up visit after two months, both cholangitis and skin lesions were healed.

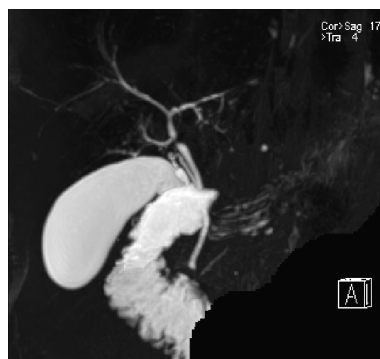
3. Discussion

The parallel course of liver abnormalities, parameters of cholestasis and psoriasis flares, as well as a negative search for any hepatotoxic drug intake, autoantibodies or viral hepatitis infection, negative history of alcohol misuse and the results of MRCP all reinforced a suspicion of neutrophilic cholangitis [1].

The outlined findings in MRCP were similar to the key MRCP features described in patients with generalized pustular psoriasis and neutrophilic cholangitis as well as in patients with neutrophilic cholangitis and psoriasis vulgaris or psoriatic arthritis [1] [4] [5].

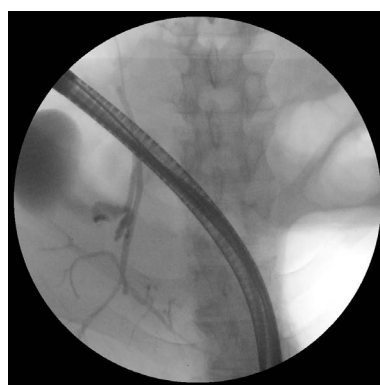
MRCP results have been recognized as acceptable criteria, although liver histology remains the gold standard for a diagnosis of neutrophilic cholangitis [1].

As described in the literature, we also found a coherence of activity of psoriasis flare and neutrophilic cholangitis [4] [5].



©KHR

Picture 1. MRCP—high-grade stricture of the principal bile duct.



©KHR

Picture 2. ERCP—restitution of the stenosis by forced administration of contrast agent.

To date no case of neutrophilic cholangitis as an elicitor of cholangiosepsis has been reported.

Neutrophilic cholangitis is often found in combination with skin diseases with intense cutaneous infiltration with polymorphonuclear leucocytes and peripheral blood neutrophilia [1].

Although the mechanism of neutrophilic infiltration of bile ducts remains speculative, enhanced expression of interleukin-8 has been documented in cholangiocytes from lesion of neutrophilic cholangitis [6], in keratinocytes from skin lesions of psoriasis vulgaris and general psoriasis pustularis [2] and synovial lesions of psoriasis arthritis [7].

In a synopsis of these findings, Interleukin-8 may play a role in the pathogenesis of neutrophilic cholangitis occurring in patients with psoriasis [1].

4. Conclusions

The range of comorbid conditions associated with psoriasis has recently grown, and the concept of psoriasis as a systemic inflammatory disorder provides the probable link with neutrophilic cholangitis.

Because of the high frequency of liver abnormalities in patients with psoriasis, a biliary participation linked with neutrophilic cholangitis should be added to the spectrum of extracutaneous manifestations of psoriasis.

References

- [1] Dieude, P., Sbidian, E., Viguier, M., Zafrani, E., *et al.* (2013) Neutrophilic Cholangitis in Psoriasis Vulgaris and Psoriatic Arthritis. *British Association of Dermatologists*, **168**, 216-218. <http://dx.doi.org/10.1111/j.1365-2133.2012.11157.x>

- [2] Marrakchi, S., Guigue, P., Renshaw, B., *et al.* (2011) Interleukin-36 Receptor Antagonist Deficiency Causes Generalized Pustular Psoriasis. *New England Journal of Medicine*, **365**, 620-628. <http://dx.doi.org/10.1056/NEJMoa1013068>
- [3] Mrowietz, U., *et al.* (2010) Comorbidity Prevalence in Psoriasis Patients: A Meta-Analysis. *Journal of the American Academy of Dermatology*, **6**. Poster Presentation at the 68th Annual Meeting of the American Academy of Dermatology, Miami Beach, FL, 5-9 March 2010, 3300.
- [4] Allez, M., Roux, M.E., Bertheau, P., *et al.* (2000) Recurrent Cholestatic Jaundice Associated with Generalized Pustular Psoriasis: Evidence for a Neutrophilic Cholangitis. *Journal of Hepatology*, **33**, 160-162. [http://dx.doi.org/10.1016/S0168-8278\(00\)80174-9](http://dx.doi.org/10.1016/S0168-8278(00)80174-9)
- [5] Viguier, M., Allez, M., Zagdanski, A.M., *et al.* (2004) High Frequency of Cholestasis in Generalized Pustular Psoriasis: Evidence for Neutrophilic Involvement of the Biliary Tract. *Hepatology*, **40**, 452-458. <http://dx.doi.org/10.1002/hep.20305>
- [6] Isse, K., Harada, K. and Nakanuma, Y. (2007) IL-8 Expression by Biliary Epithelial Cells Is Associated with Neutrophilic Infiltration in Reactive Bile Ductules. *Liver International*, **27**, 672-680. <http://dx.doi.org/10.1111/j.1478-3231.2007.01465.x>
- [7] Kane, D., Roth, J., Frosch, M., *et al.* (2003) Increased Perivascular Synovial Arthritis. *Arthritis & Rheumatology*, **48**, 1676-1685. <http://dx.doi.org/10.1002/art.10988>