

Special Article – Oral and Maxillofacial Surgery

Uncommon Infection of an Epidermoid Cyst in the Face with *Peptoniphilus harei* - A Systematic Review of the Literature Regarding Treatment Strategy

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Abstract

Purpose: Infections with *Peptoniphilus harei* are rare and no consensus about treatment exists. Based on a case report, we report on our approach and make a review of the literature. To our knowledge, we are the first to be confronted with an infected epidermoid cyst in the facial area due to *Peptoniphilus harei* in its pure form.

Material and Methods: A smear test for microbiological examination and for culture determination was taken intraoperative. *Peptoniphilus harei* could be detected by using the matrix-assisted laser desorption ionization time-of-flight mass spectrometry (MALDI-TOF) method. A literature research showed only few documentations of an infection with this microorganism.

Results: The post-operative course was uneventful. No antibiotics were needed. The microbiological examination detected *Peptoniphilus harei* as the only microorganism.

Conclusion: The clinical impact of *Peptoniphilus harei* is not clearly clarified due to poor data. Resistance cannot currently be considered as a problem. In our case report compared to the other case reports, no antibiotic therapy was initiated. For the detection of the causative microorganism, a microbiological sample is recommended.

Keywords: *Peptoniphilus harei*; *Peptostreptococcus harei*; Epidermoidcyst

Introduction

Peptostreptococcus harei was first isolated from antral lavage, from the peritoneal cavity and from a suppurative sacral ulcer. Furthermore also from abscesses of the neck area [1]. The bacterium *Peptostreptococcus* was reclassified in 2001 to the genus *Peptoniphilus*, which also includes *Peptoniphilus harei* [2]. *Peptoniphilus harei* are non-spore-forming, gram-positive, obligate anaerobic cocci. *Peptoniphilus harei* has been found mainly in polymicrobial infections [3,4]. Only in a few cases, *Peptoniphilus harei* has been isolated as the only pathogen without the involvement of other bacteria out of an infection [5-8].

In this case, report we document the first case of an infected epidermoid cyst of the face caused by *Peptoniphilus harei*. To our knowledge, we are the first to be confronted with an infected epidermoid cyst in the facial area due to *Peptoniphilus harei* in its pure form.

Case Report

A 57-year-old man introduced himself to our department. He reported that 3 weeks before a pustula on the right he manipulated cheek. After that the patient observed a slowly rising swelling on the right cheek. In the course, it came to swelling with redness and painful palpation of the soft tissues. Finally, with abscess formation in the right cheek the patient presented himself to our clinic. The patient

was in good general condition with no history of any other diseases and no history of drugs administration. In addition, the social and sexual anamnesis showed no abnormalities. The patient was afebrile and presented no ague. Because of the clear clinical findings and good general condition without further diseases, we renounced a radiological and an examination of inflammation parameters. There was no indication to take blood cultures. On the day of the presentation the abscess incision with drainage was performed. A smear test for microbiological examination and for culture determination was taken intraoperatively. After 48 hours of cultivation, gram-positive cocci were found. The detection of *Peptoniphilus harei* was achieved using the matrix-assisted laser desorption ionization time-of-flight mass spectrometry (MALDI-TOF) method. The result showed *Peptoniphilus harei* as the only microorganism. After another 24 hours, an antibiogram profile was made. The microorganism was sensitive to all the antibiotics we tested: penicillin, amoxicillin-clavulanic acid, ampicillin, clarithromycin, clindamycin and metronidazole. We did not initiate antibiotic therapy. With regular irrigation, there was a clear clinical improvement (Figure 1). During the therapy, the drainage could be removed. One week after abscess incision surgical excision of the tumorous tissue could be fulfilled (Figure 2). The histological examination revealed an epidermoid cyst without evidence of malignancy. The further course was uneventful. We could conclude the treatment two weeks after abscess incision.



Figure 1: Clear clinical improvement.



Figure 2: Surgical excision of the tumorous tissue.

Systematic Review

We aimed to identify all publications pertaining to infection with *Peptoniphilus harei* in pure culture. We used the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). Published studies were found by utilizing a thorough search strategy of the PubMed, Google Scholar and Hand search. The following search terms were used: *Peptoniphilus harei*, *Peptostreptococcus harei*, epidermoid cyst. We used the chosen articles to identify more articles that are relevant. Eligibility criterion for inclusion in the review was the detection of *Peptoniphilus harei* in pure culture.

Results and Discussion

A systematic, multilingual review of medical literature showed that only in few cases *Peptoniphilus harei* was isolated in pure culture. We examined 21 articles and identified five papers relevant to our research topic that met our criteria. The search strategy and flow diagram (Figure 3) are presented using the PRISMA guidelines.

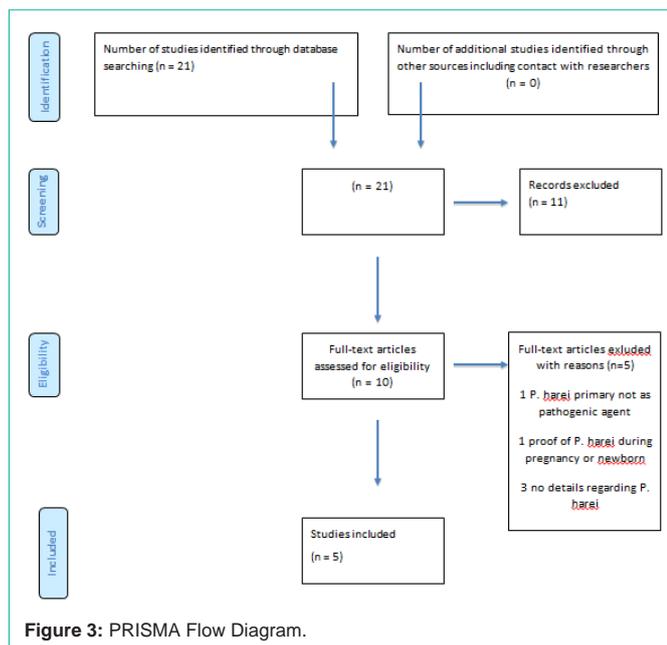
The treatment of choice for infections caused by *Peptoniphilus harei* is not established due to the lack of data. Ganiy Opeyemi Abdulrahman G.O, and Christopher Alan Gateley C.A, isolated *Peptoniphilus harei* together with *A. turicensis* from a breast abscess after piercing. Cobo et al. isolated *Peptoniphilus harei* from a purulent peritoneal inflammation in a patient with bowel obstruction in April 2016 and recently in a 60-year-old man from an infected lymphocele after radical prostatectomy. In all three cases, surgical assessment and systemic antibiotic therapy were performed [5-7]. In our case, antibiotic therapy was not required. Murdoch et al. found two strains in pure culture from abscesses in different locations. However, the therapy was not described and no statement can be made here regarding the clinical significance [1].

Peptoniphilus harei belongs to the Gram-Positive Anaerobic cocci (GPAC). The genus *Peptoniphilus* was originally described as *Peptococcus*, which was modified to *Peptostreptococcus* in 1986 [5]. In July 1997 these *Peptostreptococci* were reported by Murdoch et al. investigated further with phenotypic and genotypic methods and new subtype was isolated for the first time from antral lavage, the peritoneal cavity and a suppurative sacral ulcer. This new species of the genus *Peptostreptococcus* was then classified as *Peptoniphilus harei* [1]. The genus *Peptostreptococcus* itself was reclassified in 2001 to the genus *Peptoniphilus* [2]. *Peptoniphilus harei* occurs mainly in polymicrobial infections [3,4].

So far, only a few have been isolated in pure culture. Two strains were detected in pure culture from abscesses [1]. In a 22-year-old woman, *Peptoniphilus harei* was isolated together with *A. turicensis* from a breast abscess after piercing. However, no statement can be made here regarding the clinical significance [6]. Cobo et al. have identified *Peptoniphilus harei* from a purulent peritoneal inflammation in a patient with bowel obstruction in April 2016 and recently in a 60-year-old man from an infected lymphocele after radical prostatectomy. In both cases, the source of infection was unknown. Because *Peptoniphilus harei* belongs to the skin flora, it is assumed that the skin was responsible as the path of contamination [5,7]. We assume that our patient's skin also served as the path of contamination. Our case report describes for the first time the unique isolation of *Peptoniphilus harei* from an epidermoid cyst in the facial area. Compared to our case where the infection arose spontaneously, Cobo's et al. datum case report was preceded by surgery. Also in the second case report of Cobo et al. a patient suffering from a bilateral ovarian carcinoma was operated several times. In the described laparotomy the microbiological examination of the ascites fluid was arranged, in which the bacterium *Peptoniphilus harei* was identified [5]. It is therefore unclear whether the infection was due to one of the pre-surgery or by the immunosuppressive underlying disease.

The detection of *Peptoniphilus harei* infection passed via the establishment of a culture. Strain identification is usually done by phenotypic tests. The difficulty here is the phenotypic similarity and as well as the comparable biochemical properties of *Peptoniphilus harei* and *Peptoniphilus asaccharolyticus*. For this reason, more specific identification using molecular biological methods such as 16S rRNA sequencing is of great importance. The recent technology of Matrix-Assisted Laser Mass Spectrometry (MALDI-TOF) for the routine identification of bacteria, especially anaerobes, allows cost-effective identification of clinical isolates with high accuracy and speed [9,10]. In MALDI-TOF, the analysis molecule is added to a saturated, low-mass organic compound, also called matrix, while crystals form (co-crystallization). The analysis matrix-crystals, which are located on a metal plate, are excited with a UV laser beam with a wavelength of 337nm for a short time, so that no damage to the analysis molecule arises. Photons of the laser beam lead to sublimation and ionization of the sample to be examined. The analysis molecules and the matrix are placed into the mass spectrometer and mass spectrometry is performed [11]. To analyze the pathogens present in our smear test, we applied the system of the MALDI-TOF due to the many advantages.

An antibiotic treatment of choice for infections caused by



Peptoniphilus harej is not established due to the lack of data. Jeverica et al. demonstrated that *Peptoniphilus harej* was resistant to penicillin in 1% of cases and resistant to clindamycin even in 29% of cases. In contrast, in 0% of cases they were resistant to amoxicillin-clavulanic acid, imipenem and metronidazole [12]. Cobo et al. reported in their case a high minimal inhibitory concentration for metronidazole and can be considered resistant according to the European Committee on Antimicrobial Susceptibility Testing (EUCAST) criteria [5]. In our case, *Peptoniphilus harej* was sensitive to all the antibiotics we tested. In view of the overall situation, the resistance of *Peptoniphilus harej* cannot currently be considered as a problem. In our case, antibiotic therapy was not required. Therefore, it can be assumed that after a complete surgical removal of the infected tissue, in the absence of general symptoms, and no presence of immune deficiency an antibiotic therapy is not mandatory.

Skin infections are most commonly caused by group A beta-hemolytic streptococci and staphylococcus aureus [13]. Furthermore, aerobic and anaerobic pathogens often coexist in acute and chronic infections, as they benefit from a synergistic effect [3]. In our case, *Peptoniphilus harej* caused a cheek abscess without involvement of other bacteria and as the only pathogenic agent.

Conclusion

The clinical impact of *Peptoniphilus harej* is not clearly clarified due to poor data. Resistance cannot currently be considered as a problem. In our case report compared to the other case reports, no antibiotic therapy was initiated. For the detection of the causative microorganism, a microbiological sample is recommended.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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