Case Report

Two intriguing *Bilophila wadsworthia* cases from Hungary

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*Bilophila wadsworthia*, an obligately anaerobic Gram-negative bacillus, was first isolated from appendicitis specimens and human faeces. The two cases described here are the first report of the isolation of *B. wadsworthia* in Hungary, and include the first isolation from chronic mastoiditis complicated with a brain abscess.

**Introduction**

*Bilophila wadsworthia*, an anaerobic Gram-negative bacillus first reported in patients with gangrenous or perforated appendicitis (Baron et al., 1989), is the third most common anaerobe isolated from such patients (Baron et al., 1992a; Bennion et al., 1990; Finegold et al., 1990), usually in polymicrobial infections (Baron et al., 1989, 1992a; Finegold et al., 1992). *B. wadsworthia* requires strictly anaerobic conditions and Supplementary factors for optimal growth in *vitro* and exhibits characteristically slow colony formation (Baron et al., 1993). The lower gastrointestinal tract is the presumed ecological niche for *B. wadsworthia*, but it has occasionally been isolated from the oral cavity and vagina. It has also been isolated from various clinical specimens, e.g. from scrotal abscesses, hepatic abscesses, mandibular osteomyelitis, axillary hydradenitis suppurativa, pleural fluid, joint fluid, otitis externa, brain abscesses and blood (Baron et al., 1992b; Kasten et al., 1992), providing growing evidence that it is a significant clinical pathogen. *B. wadsworthia* is resistant to most β-lactam antibiotics, due to β-lactamase production or, possibly, to alteration of the penicillin-binding proteins, which makes this organism of considerable clinical interest. In the two cases reported here, *B. wadsworthia* was isolated during routine anaerobic culture of patient specimens. We believe that this is the first report of the isolation of *B. wadsworthia* in Hungary, and the first isolation from chronic mastoiditis complicated with a brain abscess.

**Case 1**

A 24-year-old, 20-weeks-pregnant woman was admitted with severe abdominal pain that had started about 6 h earlier. She had insulin-dependent diabetes, but had had no previous operation. She was an obese, pale woman, blood pressure 110/80 mm Hg, heart rate 80 min⁻¹ and temperature 37.7 °C, with periumbilical pain followed by nausea, and later lower right quadrant pain and vomiting. The leukocyte count was elevated, at 20.5 G/l⁻¹, with a differential count of 75% segmented neutrophils. There were no other abnormal findings. Abdominal ultrasonography indicated a normal ileocecal region and normal parenchymal organs, and the normal signs of a 20-week pregnancy. Urgent laparotomy revealed advanced appendicitis, with purulent inflammation of the surrounding tissues. The pus was aspirated and a Gram-stained smear showed many Gram-negative bacilli. Routine culture did not detect any aerobic bacteria or fungi. No aerobic bacteria were found after incubation for 48 h in an anaerobic chamber. *B. wadsworthia* grew only on BBE (Bacteroides Bile Esculin) agar alone, but was observed only after anaerobic incubation for 5 days. Histological examination revealed acute ulcerophlegmonous appendicitis. The post-operative period was uneventful and the patient was discharged on the fourth post-operative day in good condition with her pregnancy intact.

**Case 2**

A 44-year-old woman was admitted to the intensive care unit with suspected meningitis. She had a 2-week history of acute suppurative otitis media, which had been treated with amoxicillin. She complained of a severe right, dull occipital headache and dizziness, with nausea and vomiting. She was febrile (38.9 °C), assumed a forced position in bed (stiff neck), and had focal neurological symptoms: headache, sensitivity to light, nausea, otorrhoea and right otalgia. She had a leukocytosis and an elevated erythrocyte sedimentation rate. The mastoid radiographs were characteristic of chronic mastoiditis: cloudiness of the mastoid air cells, with fuzziness of the bony partitions. The presence of right-sided extradural brain abscesses was demonstrated by computerized tomography (CT), and a CT scan of the temporal bones revealed soft tissue changes within the middle ear and mastoid. The patient underwent burr hole drainage and irrigation of the extradural abscesses, followed by a simple right mastoidectomy and Penrose drainage of the mastoid cavity. Intraoperative cultures grew *Pseudomonas aeruginosa* and *Enterococcus faecalis* after aerobic incubation, and a mixed
anaerobic flora which contained *Prevotella intermedia*, *Prevotella buccae*, *Peptostreptococcus magnus* (*Finegoldia magna*), *Peptostreptococcus micros* (*Micromonas micros*), *Prevotella loescheii* and *B. wadsworthia*. *B. wadsworthia* grew only after incubation for 4 days on BBE agar in an anaerobic chamber. The patient was treated with meropenem for 2 weeks and then with oral amoxycillin-clavulanic acid for a further 2 weeks. She was discharged on post-operative day 14. The treatment achieved complete cure and full recovery of the mastoiditis and no recurrence of abscesses.

**Discussion**

*B. wadsworthia* was named by Baron *et al.* (1989) after its isolation from approximately half of all anaerobic cultures performed on specimens from patients with perforated or gangrenous appendicitis. It was later also isolated from different types of infections after prolonged anaerobic incubation. *B. wadsworthia* is a bile-tolerant, strongly catalase-positive, non-motile, indole-negative, non-spore-forming, obligately anaerobic Gram-negative bacillus which is usually urease-positive (Jousimies-Somer *et al.*, 2002). Major amounts of acetic acid and minor amounts of succinic acid are produced from glucose metabolism (Baron, 1997). It was originally thought to be resistant to antimicrobial agents generally used to treat anaerobic infections, because of the difficulty in interpretation of agar dilution susceptibility (Baron *et al.*, 1993); the results of conventional susceptibility tests, which rely on adequate growth in a standardized medium, are difficult to interpret with anaerobic species (Citron *et al.*, 1991). Results obtained with standardized methods may overestimate its resistance, and initial susceptibility studies suggested that *B. wadsworthia* was resistant to imipenem, cefoxitin, penicillin G and other β-lactam antibiotics (Summanen *et al.*, 1992). Although β-lactamase production was not demonstrated in an earlier study (Baron *et al.*, 1989), it was later reported that 87% of clinical isolates of *B. wadsworthia* were β-lactamase-positive (Baron, 1997). The two isolates described here exhibited typical phenotypic characteristics (they were strongly catalase- and urease-positive, nitrate-positive, indole-negative and asaccharolytic by the bioMérieux API 20A test). GLC analysis (Shimatzu) showed a major amount of acetic acid and a minor amount of succinic acid. In both cases, *B. wadsworthia* was detected on BBE agar only after incubation for 4–5 days. It appeared as tiny colonies that were circular, black or translucent with a dark centre. In a Gram-stained smear the cells were Gram-negative and pleomorphic. The isolates were sensitive to kanamycin and colistin and resistant to vancomycin with the identification disks. Antibiotic susceptibilities were determined with the Etest (AB Biodisk) according to the manufacturer’s instructions. The isolate from the first patient did not produce β-lactamase by the nitrocephin method, but was resistant to penicillin G in the Etest (MIC = 16 mg l⁻¹). The isolate from the second patient produced β-lactamase and had a penicillin G MIC of ≥32 mg l⁻¹. Both were susceptible to clindamycin, amoxy-

cillin-clavulanic acid, metronidazole, imipenem and chloramphenicol, with very low MICs. *B. wadsworthia* has been isolated in specimens from patients with appendicitis usually as part of a mixed flora, but in our patient with acute appendicitis, *B. wadsworthia* was the only pathogen isolated. Only two previous examples of otological infections due to *B. wadsworthia* have been reported: a patient with post-operative otitis externa following stapedectomy (Schumacher & Bücheler, 1997) and a patient with chronic otitis media complicated with a brain abscess (Marina *et al.*, 1997). Because *B. wadsworthia* has been recognized only relatively recently, many clinical microbiologists may not be familiar with its isolation and characterization and it is easily overlooked because of its slow and tiny growth. Microbiologists should be alert to need to incubate the anaerobic cultures for long enough and to examine the plates closely to detect these organisms in complex microbiological cultures.

**Acknowledgements**

This work was supported by grant T032385 from the Hungarian National Research Foundation (OTKA).

**References**


