covered by a thin layer of flattened epithelial cells. Cutaneous appendages are absent or rudimentary in the lesions and may be malformed or calcified in the surrounding areas.[5] Only rarely is a biopsy necessary since the diagnosis is usually obvious from clinical examination.

If the diagnosis is missed at birth, the scarring may later be confused with morphea, discoid lupus erythematosus, epidermal nevus and nevus sebaceous. However, the small lesions heal spontaneously, with the formation of an atrophic or hypertrophic scar, although keloid formation can occur. Any infection should be treated with appropriate antibiotic agents. The larger lesions should be closed with plastic surgery.

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Infective endocarditis due to *Leptotrichia buccalis*: a case report

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A patient with Down's syndrome presented with infective endocarditis due to *Leptotrichia buccalis*. The source of the infection was not detected, but the predisposing factor was a complex cardiac malformation. The disease followed a subacute course, had a number of immunologic manifestations and was successfully treated with a 28-day course of penicillin G, given intravenously. *L. buccalis* has never been reported before as a cause of endocarditis.

Un patient atteint du syndrome de Down a été vu souffrant d'une endocardite infectieuse à *Leptotrichia buccalis*. La source de l'infection n'a pu être décelée, mais le facteur prédisposant était une malformation cardiaque complexe. La maladie a eu une évolution subaiguë, avec un certain nombre de manifestations immunologiques; elle fut traitée avec succès par un traitement de 28 jours à la pénicilline G par voie intraveineuse. *L. buccalis* n'a jamais été signalé auparavant comme cause d'une endocardite.

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Infective endocarditis is rarely caused by anaerobic gram-negative bacteria; few cases have been reported.[1,2] One of these bacteria, *Leptotrichia buccalis*, is a normal constituent of the oral flora.[3-5] It has infrequently been associated with infections in humans,[5] the first case of systemic infection having been reported only recently.[6] We report the first case of infective endocarditis due to this microorganism.

Case report

Clinical course

A 24-year-old man with Down's syndrome was admitted to another hospital for evaluation of a flu-like illness of 8 weeks' duration. At that time he had a grade 4/6 holosystolic murmur at the apex, splenomegaly and two episodes of hemoptysis. Blood tests showed the following results: hemoglobin level 60 g/L, leukocyte count 4.5 × 10^9/L (72% neutrophils and 20% band forms), erythrocyte sedimentation rate (ESR) 160 mm/h and platelet count 153 × 10^9/L. Testing for rheumatoid factor by latex agglutination, the direct Coombs' test and the antistreptolysin O test all gave positive results. The heterophil antibody test results, however, were negative. The serum concentration of creatinine was 2.5 mg/dL (221 μmol/L) and of the third and fourth components of complement 0.48 and 0.09 g/L respectively. Three blood samples drawn over a 24-hour period were positive for gram-negative anaerobic bacteria when cultured. There was proteinuria and microscopic hematuria. An echocardiogram showed vegetations on the anterior leaflet of the mitral valve.

The patient was then transferred to our institution with a presumptive diagnosis of infective endocarditis. He was pale but in no distress. The grade 4/6 holosystolic murmur was heard at the third and fourth inter-

costal spaces on the left of the sternum and was widely transmitted over the precordium. There were no peripheral cutaneous signs of endocarditis. Hepatosplenomegaly was present, but the results of the rest of the physical examination were unremarkable. The patient stated that he had last visited a dentist more than a year earlier.

The results of our laboratory studies were similar to those obtained in the first hospital. The serum levels of electrolytes were normal, of creatinine 4.3 mg/dL (380 μmol/L) and of iron 7 μg/dL (1.2 μmol/L). The urine sediment contained 40 to 50 erythrocytes and 0 to 2 leukocytes per high-power field; the results of urine culture were negative. Serum protein electrophoresis showed an increased γ-globulin concentration (35 g/L) and a low level of albumin (27 g/L). The serum level of IgG was 20.48 g/L, of IgM 4.03 g/L and of IgA 1.44 g/L. Cryoglobulins were not found. Chest x-ray films, an electrocardiogram and a pulmonary technetium 99m scan all showed no abnormalities. The echocardiogram, however, again showed vegetations on the anterior leaflet of the mitral valve. Five blood samples drawn over a 48-hour period were positive for gram-negative anaerobic bacteria. A presumptive diagnosis of *Fusobacterium* sp. endocarditis was made and the patient treated with penicillin G, 10×10^6 U/24 h in a continuous intravenous infusion for 28 days.

The course of the disease in hospital was uneventful. Blood samples obtained 1 week after the beginning and 1 week after the end of treatment were sterile.

Initial in-vitro studies showed that the bacterium was quite susceptible to penicillin G, but we were unable to perform the tests for serum bactericidal activity. At the time of discharge the patient's hemoglobin level was 93 g/L, the hematocrit 27.6%, the platelet count 257×10^9/L, the ESR 60 mm/h and the serum creatinine concentration 4.7 mg/dL (416 μmol/L). The urine showed 10 to 30 erythrocytes per high-power field.

At a follow-up assessment 5 months later the patient was doing well. The cardiac sounds were unchanged, but the hepatosplenomegaly had disappeared. The laboratory values were: hematocrit 40.2%, hemoglobin level 136 g/L, leukocyte count 4.6×10^9/L, serum creatinine concentration 1.6 mg/dL (141 μmol/L) and urea nitrogen level 19 mg/dL (urea level 6.8 mmol/L). The ESR was 43 mm/h, the results of rheumatoid factor and Coombs' tests were negative, the concentration of the third and fourth components of complement and the results of serum protein electrophoresis were normal, and there were 0 to 4 erythrocytes per high-power field in the urine sediment. Cardiac catheterization revealed an endocardial cushion defect and coarctation of the aorta.

Bacteriologic findings

Blood samples cultured with the Bactec 460 system (Johnston Laboratories, Inc., Cockeysville, Maryland) were positive in 24 to 48 hours. Gram staining them showed long, thin bacilli with pointed ends. Slight growth was obtained on chocolate agar after 48 hours in a 5% concentration of carbon dioxide at 35°C, but much better growth was observed on trypticase soy agar supplemented with vitamin K, hemin and sheep blood incubated anaerobically in a GasPak jar (Baltimore Biological Laboratories, Cockeysville, Md.) at 35°C. On the latter medium two types of colonies were seen, one smooth and the other rough. Gram staining of these subcultures showed both the long, thin bacilli seen initially and bacilli containing granulations, which gave them a diphtheroid appearance. Use of chromatogenic cephalosporin substrate showed no β-lactamase. The bacilli could not be identified with the API 20A system (Analytab Products, Inc., Plainview, New York).

The unknown strain was sent to the bureau of bacteriology at the Laboratory Centre for Disease Control in Ottawa, where it was identified as *L. buccalis*. Gas chromatography after growth on peptone–yeast–glucose agar revealed lactic acid, formic acid and acetic acid. The organism fermented amygdalin, cellobiose, fructose, glucose, lactose, maltose, mannose, raffinose, salicin, sucrose, trehalose and xylose but not arabinose, erythritol, glycogen, inositol, mannitol, melezitose, melibiose, rhamnose, ribose or sorbitol. Esculin was hydrolysed and milk was curdled, but starch was not acidified, gelatin was not liquified, neither indole nor catalase was produced, and nitrate was not reduced.

The in-vitro susceptibility of the bacterium was determined at the Centers for Disease Control in Atlanta, Georgia (Table I). The method used was broth microdilution, in which a brain–heart infusion broth enriched with vitamin K_1 (0.5 μg/mL) and hemin (5 μg/mL) was inoculated with 10^5 colony-forming units per millilitre and incubated for 48 hours in an anaerobic chamber.

Table I—Susceptibility of one isolate of *Leptotrichia buccalis* to 25 antimicrobial agents

Antibiotic	Minimum inhibitory concentration (μg/mL)
Penicillin G	≤ 0.06
Sch 29482	≤ 0.06
Aztreonam	2
Azlocillin	≤ 0.12
Mezlocillin	≤ 0.12
Piperacillin	≤ 0.25
Carbenicillin	0.5
Cefoxitin	≤ 0.06
Cefotetan	0.12
Cefotaxime	≤ 0.06
Moxalactam	1.0
Cefoperazone	≤ 0.06
Ceftriaxone	≤ 0.06
Cefmenoxime	≤ 0.12
Ceftazidime	1.0
Ceftizoxime	≤ 0.5
Doxycycline	≤ 1.0
Chloramphenicol	4
Metronidazole	1.0
Erythromycin	4
Clindamycin	≤ 0.015
Pirlimycin	0.03
Rifampin	2
Thienamycin	0.12
Trimethoprim/ sulfamethoxazole	≤ 0.015/0.3

Discussion

L. buccalis is a gram-negative, non-spore-forming anaerobic bacterium in the family of Bacteroidaceae. This family includes three genera: *Bacteroides*, *Fusobacterium* and *Leptotrichia*. *L. buccalis* is the only species recognized in the last genus.[5,6]

L. buccalis has been recovered from the oral cavity, the intestine

and the vagina of humans.[4,5] Infections due to *L. buccalis* have been reported very infrequently, though, and in almost all cases the organism was part of a mixed culture of aerobic and anaerobic bacteria. These infections were usually localized in the mouth or the gastrointestinal tract.[7,8] Asymptomatic bacteremia following dental extraction or manipulation has also been reported.[9,10] Only one case of clinically manifest bacteremia has been reported, in a patient who had chronic lymphocytic leukemia and was receiving chemotherapy; the source of the bacteria was believed to be a lung abscess.[6]

The clinical picture in our patient was consistent with a diagnosis of infective endocarditis. He had sustained bacteremia, with eight blood cultures being positive over a period of 2 weeks, vegetations on the mitral valve, detected by echocardiography, and a debilitating course of 2 months' duration that included pulmonary embolism, splenomegaly, anemia and immunologic manifestations, which resolved after treatment.

Endocarditis due to gram-negative anaerobic bacteria generally follows a subacute course.[2] In most cases an underlying cardiac disease is present; systemic embolism is the commonest complication. Immunologic manifestations have not been well described. Our patient presented several transient immunologic changes: positive results of rheumatoid factor and Coombs' tests, and immune-complex nephritis, with a low level of complement, a very high level of γ-globulin (35 g/L) and increased concentrations of IgG and IgM in the serum. The very strong immune response known to occur with *L. buccalis* in the human probably explains these findings.[11]

Our patient's infection responded well to penicillin G, as would be expected from the microorganism's extreme sensitivity to that antibiotic.

We think that we have documented the first case of endocarditis due to *L. buccalis*. This disease occurred in a patient whose immune system was not suppressed, and it followed a subacute course, was associated with a number of immunologic abnormalities and was successfully treated with a 28-day course of penicillin G. The source of the infection was not discovered but was likely in the buccal cavity. The predisposing factor was a complex congenital cardiac malformation. *L. buccalis* has the potential to cause serious disease.

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Scheduling work-ins

In most practices, patients who must be seen the same day they call are the number one scheduling problem. This can be solved by taking a systematic approach to developing a work-in schedule. For eight weeks, enter work-in patients with a different colour in the appointment book. Then calculate the average number for each day of the week. If you leave approximately that percentage of open time per day, your work-in schedule will be specifically attuned to your practice.