Frontal osteomyelitis (Pott's puffy tumour) associated with *Pasteurella multocida* – A case report and review of the literature

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R Skomro, KL McClean. Frontal osteomyelitis (Pott's puffy tumour) associated with *Pasteurella multocida* – A case report and review of the literature. Can J Infect Dis 1998;9(2):115-121.

A 58-year-old woman presented with progressive midforehead swelling and erythema with frontal headache. Investigations revealed erosion of the anterior wall of the frontal sinus with subgaleal abscess formation, establishing a diagnosis of Pott's puffy tumour. *Pasteurella multocida* was isolated in pure growth from an aspirate of the abscess. *P multocida* is a rare cause of sinusitis. It is isolated from the respiratory tract of asymptomatic individuals and, more commonly, patients with chronic respiratory conditions. Although a cause of osteomyelitis associated with animal bites or scratches, *P multocida* has not previously been implicated as a cause of frontal osteomyelitis or Pott's puffy tumour. A review of reported cases of Pott's puffy tumour, including clinical presentation, microbiology, treatment and outcome, is provided.

Key Words: *Frontal osteomyelitis,* Pasteurella multocida*, Pott's puffy tumour, Sinusitis*

Ostéomyélite frontale (tumeur de Pott) associée à *Pasteurella multocida* : Rapport de cas et survol de la littérature

RÉSUMÉ : Une femme de 58 ans s'est présentée avec une enflure progressive au milieu du front, accompagnée d'un érythème et de céphalée frontales. Les épreuves de laboratoire ont révélé la présence d'une érosion de la paroi antérieure du sinus frontal avec abcès sous-galéal en formation, ce qui a confirmé le diagnostic de tumeur de Pott. *Pasteurella multocida* a été isolé dans une croissance pure à partir d'un aspirat de l'abcès. *P. multocida* est une rare cause de sinusite. On l'isole dans les voies respiratoires de sujets asymptomatiques et, plus fréquemment, de patients atteints de maladies respiratoires chroniques. *P. multocida* est une cause d'ostéomyélite associée aux morsures ou aux égratignures infligées par des animaux, et n'avait jamais auparavant été incriminé dans l'ostéomyélite frontale ou tumeur de Pott. Un survol des cas de tumeur de Pott signalés, y compris le tableau clinique, la microbiologie, le traitement et l'issue du traitement est présenté ici.

Sir Percival Pott, whose name is associated with such entities as Pott's puffy tumour (PPT), Pott's fracture and Pott's disease, was one of the leading surgeons of the 18th century. In 1760 he described a "puffy, circumscribed, indolent tumour of the scalp and the spontaneous separation of the pericranium from the scalp under such tumour" associated with an underlying osteomyelitis of the frontal bone, a condition now known as PPT (1). Pott's original case developed following head trauma. In 1775 he described a second case complicating frontal sinusitis.

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Received for publication Feb 6, 1997. Accepted May 1, 1997

Infection spreads from the sinus through the diploic veins anteriorly to the subgaleal space, resulting in frontal bone osteomyelitis, erosion through the frontal bone and subperiosteal abscess formation. Posterior spread occurs by direct extension through areas of osteomyelitis in the posterior wall of the sinus and may involve underlying dura with formation of extradural empyema. Bacterial spread along small vessels traversing the dura may result in subdural empyema, an inflammatory reaction in the subarachnoid space, or involvement of brain parenchyma. Intracranial spread is facilitated by the absence of valves in the diploic venous system that provide direct communication among sinus mucosa, marrow spaces of the frontal bone and dural veins.

Intracranial complications of acute frontal sinusitis, though declining in incidence since the onset of the antibiotic era, have been reported in 10% of patients hospitalized for treatment of frontal sinusitis (2). The reported incidence of intracranial complications is higher in patients presenting with PPT. In a review of 11 cases of PPT by Feder et al (3) eight of 11 patients suffered intracranial complications.

We describe a case of PPT associated with chronic sinusitis and present a literature review of PPT cases.

CASE PRESENTATION

A 58-year-old woman was admitted to Royal University Hospital, Saskatoon, Saskatchewan in April 1995 with frontal cellulitis. She had been well until three weeks before admission when she noted a raised, red lesion in the centre of her forehead. Twenty-four hours after the appearance of the lesion, she sought medical advice and was prescribed topical corticosteroids for nonspecific dermatitis. After three days of topical corticosteroid therapy, she again sought medical advice because of progressive enlargement of the lesion. A diagnosis of cellulitis was made, and oral cephalexin, 500 mg qid, was prescribed. During the course of therapy with cephalexin, she consulted a dermatologist because of continued worsening of the lesion. A plastic surgery consultation was obtained five days later. By this time, the lesion had spread to involve most of the forehead with extension of swelling and erythema onto the bridge of the nose and to both periorbital areas. She denied fevers, chills or sweats. She had no history of antecedent trauma, respiratory infection or acute sinusitis. She had a long history of frontotemporal headaches recently diagnosed as migraines. Progressive worsening of the headaches had been noted for several months before the appearance of the frontal lesion. There was a history of recurrent sinusitis dating back 20 years and previous maxillary sinus irrigation. Her only exposure to animals was an occasional visit to a family member who owned a dog. There was no history of bites or scratches from the dog, or extensive handling of the dog.

On physical examination, she was alert and oriented, with a temperature of 36.9°C, blood pressure of 115/64 mmHg, heart rate of 80 beats/min and a respiratory rate of 14 breaths/min. There was a large area of swelling and erythema on the forehead with extension onto the nasal bridge and left periorbital area (Figure 1). The lesion was exquisitely tender, warm and fluctuant. The maxillary sinuses were nontender. The remainder of her physical examination, including neurological examination, was unremarkable. The leukocyte count was 5.0×10^9/L with 3.7×10^9/L neutrophils, 0.9×10^9/L lymphocytes and 0.3×10^9/L monocytes. Hemoglobin was 128 g/L, platelet count was 311×10^9/L and erythrocyte sedimentation rate (ESR) was 15 mm/h.

Incision and drainage of the fluctuant area yielded purulent material with moderate white blood cells and very rare Gram-negative bacilli on Gram stain. X-rays of the paranasal sinuses showed normal maxillary sinuses and thick-walled frontal sinuses with the right frontal sinus markedly smaller than the left. The ethmoid and sphenoid sinuses were described as "somewhat cloudy". A contrast-enhanced computed tomography (CT) scan demonstrated bilateral nonuniform thickening in the maxillary and ethmoid sinuses, a small defect in the medial wall of the right maxillary sinus consistent with her previous surgery, and opacification of the frontal sinuses consistent with sinusitis. There was a defect in the anterior wall of the left frontal sinus immediately adjacent to the midline, contiguous with the area of soft tissue swelling. A large defect was present in the posterior wall of the left frontal sinus with only a thin enhancing membrane separating sinus contents from the frontal lobe (Figure 2). Increased sclerosis of bony margins suggested chronic osteomyelitis. A small, nonenhancing fluid collection with minimal mass effect was present along the anterolateral aspect of the left frontal lobe.

Broad spectrum empiric antibiotic therapy with ciprofloxacin 400 mg intravenously bid, ceftazidime 2 g intravenously every 8 h and metronidazole 1 g intravenously every 6 h was commenced. The wound was managed with saline gauze packing. A neurosurgical consultant felt that the subdural effusion represented a sympathetic effusion and drainage was not indicated.

Cultures of the drainage yielded pure growth of *Pasteurella multocida*. However, because prior antibiotic therapy may have impaired recovery of associated organisms, she was continued on broad spectrum therapy with cefotaxime, 2 g intravenously every 8 h and 500 mg orally tid. Following a week of in-hospital intravenous antibiotics, she was discharged home to continue intravenous cefotaxime and oral clindamycin for four weeks. This was followed by oral penicillin therapy, 600 mg qid. Because free drainage of the sinus was occurring through the bony defect, immediate sinus drainage was not performed. Obliteration of the sinus and a reconstructive procedure was planned following resolution of the osteomyelitis. Opacification of the sinus persisted despite continued oral penicillin for five months and medical therapy with decongestants and intranasal steroids. Operative exploration of the sinus demonstrated acute purulent sinusitis with a small amount of necrotic bone. Aerobic and anaerobic cultures of necrotic debris from the sinus did not yield any organisms. No specimens were sent for pathological examination. The frontal nasal duct was completely obstructed by scar tissue. Surgical management included debridement of all necrotic material and the hyperplastic sinus mucosa, a left Lynch procedure, reconstruction of the frontal nasal duct, bilateral subraucous re-

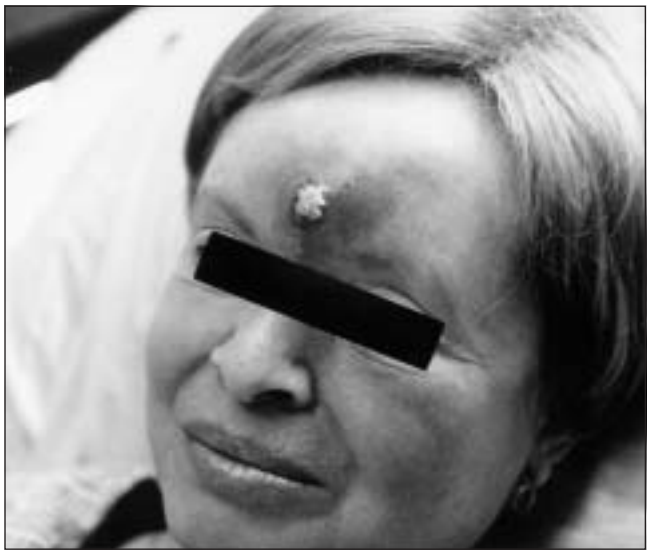

Figure 1) *Midfrontal lesion, Pott's puffy tumour (postincision and drainage). Normal saline packing is visible in the centre of the lesion*

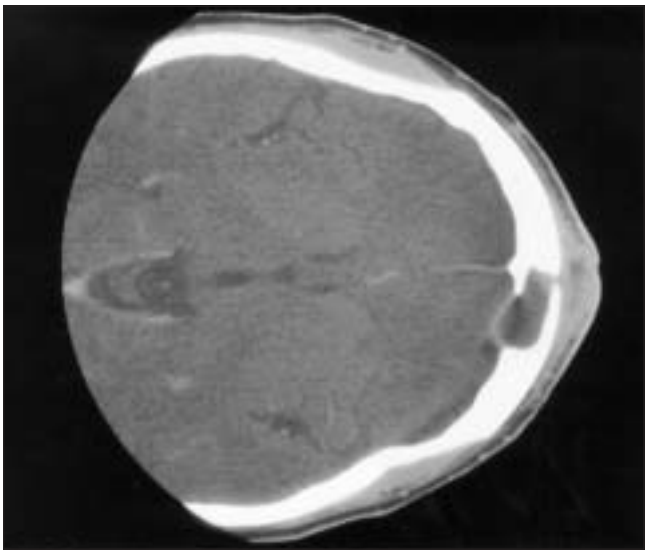

Figure 2) *Contrast-enhanced computed tomography scan demonstrating subgaleal abscess, erosion of anterior and posterior wall of left frontal sinus and small epidural fluid collection*

section of the inferior turbinates and bilateral nasal antrostomies. Postoperative recovery was uneventful.

LITERATURE REVIEW

A MEDLINE search using the terms "Pott's puffy tumor", "frontal osteomyelitis" and "frontal sinusitis" and a review of references yielded 53 cases of PPT described in the English literature since 1945 (Table 1). All patients diagnosed as PPT by the authors as well as cases of frontal osteomyelitis with subgaleal abscess found on CT scan or at surgery are included.

Cases of PPT occurred in all age groups, with patients ranging in age from two to 83 years. The disease predominantly affects young males – a male to female ratio of 9:1 – with 70% of patients under 30 years of age. Sinusitis, head trauma or cranial surgery was a predisposing factor in 75% of cases. Upper respiratory tract infection (other than sinusitis), intranasal cocaine use (with associated sinusitis) and allergic rhinitis preceded the development of PPT in a minority of patients. Trauma, as a predisposing factor, was more common in adults than children (46% versus 14%), and sinusitis was more common in children (52% versus 33%). The reason for the striking male predominance is unclear. All cases following trauma occurred in male patients but this accounts for only 10 of 54 cases. The greater frequency of respiratory disease in male children may contribute to the male predominance.

Frontal sinusitis is often clinically silent and may persist unrecognized for prolonged periods. Symptoms, when present, may be nonspecific, and include headache, nasal discharge and fever. Even in the presence of PPT, fever may be absent. In our review, temperature was reported in only 35 cases, 13 (37%) of which were afebrile. Feder et al (3) reported fever in only five of 11 cases.

Neither leukocytosis (white blood cell count 9.5×10^9 or greater) nor elevated ESR provided a reliable indication of the presence of underlying osteomyelitis in patients presenting with frontal cellulitis or sinusitis. When reported, leukocytosis was absent in 14% of children and 50% of adults (22 cases). ESR was reported in only 14 cases and was normal in two cases. In a review of cases of osteomyelitis of the skull only one of nine patients with osteomyelitis of the skull had an elevated ESR (4).

Gram stain of the subgaleal abscess or sinus aspirate can be helpful in directing initial antibiotic therapy; however, Gram stain results were infrequently documented. Results of bacterial cultures were available in 36 cases. A single pathogen was isolated in 53%, multiple organisms in 47%. Four cultures yielded no growth. *Streptococcus* species and *Staphylococcus aureus* were the most common isolates in 17 (47%) and eight (22%) patients, respectively. Anaerobic bacteria were isolated from 10 (28%) cases.

The antibiotic regimens used, mode of administration and duration of therapy were highly variable. In most reports two or more antibiotics were used, usually achieving broad spectrum activity with agents capable of penetrating the central nervous system. The marked variability in antibiotic therapy precludes conclusions about the efficacy of various regimens. In most cases, therapy was continued for four to six weeks.

Surgical intervention was the only therapeutic option for patients with frontal osteomyelitis in the pre-antibiotic era and remains a crucial component of the care of patients with PPT. The extent and timing of surgical intervention is dictated by the presence of intracranial complications and necrotic bone, and the response to antibiotic therapy. When craniotomy is indicated, subperiosteal abscesses and affected sinuses should be drained at the time of craniotomy. In the absence of intracranial complications, most patients in this review were managed by incision and drainage of the subgaleal abscess followed by sinus trephination. In our patient, failure to achieve definitive sinus drainage led to persistent inflammation of the sinus despite prolonged treatment with antibiotics, decongestants and nasal steroids. Unrecognized complete obstruction of the frontal nasal duct likely accounted

TABLE 1
Review of cases of Pott's puffy tumour

Case (reference)	Age (years)/ Sex	Associated conditions	Culture results	Complications	Outcome
1 (32)	26/M	Craniotomy	NR	Recurrent subperiosteal abscess	Recurrence
2 (32)	42/M	Craniotomy	NR	NR	Full recovery
3 (32)	44/M	Craniotomy	NR	NR	Full recovery
4 (32)	47/M	Craniotomy	NR	NR	Full recovery
5 (6)	6/M	Trauma	NR	None	NR
6 (7)	16/M	Trauma	Bacteroides, beta-hemolytic streptococci, *Enterococcus faecalis*, *Eschericia coli*, peptostreptococcus	Epidural/frontal abscess	Full recovery
7 (7)	14/M	Trauma	Beta-hemolytic streptococcus	Epidural abscess	Full recovery
8 (17)	28/M	Remote trauma	Coagulase negative staphylococcus, *Haemophilus* species	None	Full recovery
9 (18)	27/M	Frontal sinus injury	NR	NR	Full recovery
10 (19)	40/M	Trauma	NR	Orbital abscess	Full recovery
11 (33)	83/M	Trauma	*Haemophilus influenzae*, nontypeable	None	Full recovery
12 (23)	26/M	Trauma	*Salmonella typhi* type B	Epidural empyema	Full recovery
13 (34)	2/M	Trauma, chronic granulomatous disease	*Serratia marcesens*, *Staphylococcus epidermidis*	None	NR
14 (17)	36/M	Trauma, sinusitis	*Staphylococcus aureus*	Recurrences	Full recovery
15 (32)	52/M	Craniotomy, sinusitis	NR	NR	Full recovery
16 (3)	6/F	Sinusitis	*Peptostreptococcus* species, *Bacteroides melaniongenicus*	None	Full recovery
17 (35)	14/M	Sinusitis	*S aureus*	None	Full recovery
18 (19)	7/M	Sinusitis	NR	Extradural abscess	Full recovery
19 (21)	14/M	Sinusitis	*S aureus*	Epidural abscess	Full recovery
20 (3)	12/F	Sinusitis	Alpha-hemolytic streptococcus	Epidural, frontal abscess	Seizures
21 (6)	15/M	Sinusitis	*Streptococcus milleri*	Frontal/subdural abscess	Blind spot enlarged
22 (36)	13/M	Sinusitis	*S aureus*, viridans group streptococcus	None	Full recovery
23 (36)	17/M	Sinusitis	Coagulase-negative staphylococcus	Epidural/intraorbital extension	Full recovery
24 (10)	17/M	Sinusitis	Nonhemolytic streptococcus	Subdural abscess, seizures	Left hemiparesis
25 (11)	39/M	Sinusitis	No growth	Epidural abscess	NR
26 (8)	19/M	Sinusitis	No growth	Sub/epidural empyema	Full recovery
27 (8)	13/F	Sinusitis	Anaerobic streptococcus	Increased intracranial pressure	Full recovery
28 (9)	7/M	Sinusitis	*S aureus*	Orbital abscess, seizures	Full recovery
29 (14)	25/M	Sinusitis	Mixed *Streptococcus* species	Sub/epidural abscess, seizures	Seizures
30 (37)	16/M	Sinusitis	Beta-hemolytic streptococcus, *Bacteroides fragilis*	Brain abscess	Full recovery
31 (17)	59/M	Sinusitis	Alpha-hemolytic streptococcus, *Hemophilus* species	Frontal cerebritis	Full recovery
32 (16)	16/M	Sinusitis	Alpha-hemolytic streptococcus, *H influenzae*	Epidural abscess	Full recovery

Continued on next page

TABLE 1 continued
Review of cases of Pott's puffy tumour

Case (Reference)	Age (year)/ Sex	Associated conditions	Culture results	Complications	Outcome
33 (14)	14/M	Sinusitis	Alpha-hemolytic streptococcus	Subdural empyema	Full recovery
34 (15)	14/M	Sinusitis	*Bacteroides* species	Epidural abscess	Full recovery
35 (present case)	58/F	Sinusitis	*Pasturella multocida*	None	Full recovery
36 (38)	34/M	Intranasal cocaine	Group A streptococcus, mixed anaerobes, propionibacterium	None	Full recovery
37 (7)	18/M	URTI	*Streptococcus constellatus*, *B fragilis*, peptostreptococcus, propionibacterium	Multiple abscesses	Full recovery
38 (6)	14/M	URTI	Mixed *Streptococcus* species	Subdural empyema	Full recovery
39 (17)	14/M	URTI	*Streptococcus intermedius*, *B fragilis*, peptostretococcus	Subdural empyema, brain abscess	Seizures
40 (20)	13/M	URTI	*Eikenella corrodens*, *Proteus mirabilis*	Orbital cellulitis, epidural abscess	Full recovery
41 (7)	15/M	URTI	*S aureus*, *Peptococcus magnus*, propionibacterium acnes	Epidural and cerebral abscess	Full recovery
42 (20)	26/M	Nasal polyps	NR	None	Full recovery
43 (20)	36/M	Nasal discharge	Alpha-hemolytic streptococcus, mixed anaerobes	None	Full recovery
44 (39)	19/M	Allergic rhinitism	Alpha-hemolytic streptococcus, fungus	None	Full recovery
45 (12)	54/F	None	No growth	Orbital erosion	Full recovery
46 (5)	13/M	None	no growth	Epidural abscess	Full recovery
47 (8)	15/M	None	*S aureus*	Orbital abscess, coma	Full recovery
48 (40)	72/M	None	*S aureus*	None	Chronic osteomyelitis
49 (41)	26/M	None	*H influenzae* type A	None	Full recovery
50 (13)	10/M	NR	NR	NR	NR
51 (13)	13/M	NR	NR	Frontal abscess	NR
52 (13)	11/M	NR	NR	Pericranial, subdural abscess	NR
53 (22)	22/M	NR	*E corrodens*	Subdural empyema	Full recovery
54 (42)	NR	NR	NR	NR	NR

NR Not reported; URTI Upper respiratory tract infection

for the failure of conservative management. Adequate long term sinus drainage, in addition to drainage of the subgaleal abscess, is an essential component of therapy.

Intracranial complications associated with PPT include subdural or epidural empyema, meningitis, cerebritis and cerebral abscess (3,5-23). Extracranial complications result from the extension of the infection into the periorbital tissues, orbit or subgaleal space. In this review the overall complication rate was 61% with intracranial complications occurring in 52%, undoubtedly reflecting a degree of reporting bias in favour of severe or complicated cases. Complications were more frequent in children (83%) than adults (42%). Intracranial complications occurred in 75% of children and 25% of adults; epidural and subdural abscesses were most common, affecting 33% and 18% of the patients, respectively.

Before the introduction of antibiotics, the mortality of frontal osteomyelitis was as high as 60% (24). In the antibiotic era, Bordley and Bichofberger (24) described a series of 28 patients with frontal osteomyelitis seen between 1952 and 1965 and reported one death (3.5%). Similar case series for PPT are not available. No deaths were seen among the 46 patients in whom outcome was reported in this review. Three patients suffered from seizures, one from hemiparesis and two from chronic osteomyelitis. The remaining patients made a complete recovery.

DISCUSSION

The patient described in this report is the second case of PPT in an adult female in the English literature during the antibiotic era and the first reported case due to *P multocida*. Our patient presented with progressive midforehead swelling and frontal headaches but without systemic features of infection. Ten of the 25 adult cases identified in this review presented in a similar manner with isolated frontal headaches and swel-

ling. The absence of fever, leukocytosis and elevated ESR is not unusual and may contribute to delay in recognition of underlying disease when a patient presents with frontal cellulitis.

Culture of the abscess aspirate from our patient unexpectedly yielded pure growth of *P multocida*, a small Gram-negative coccobacillus, best known for its role in infections of skin and soft tissues resulting from animal bite wounds. The organism inhabits the digestive tracts of many animals, particularly cats and dogs (25). A review of 446 cases of human *P multocida* infections found that 48% involved skin or subcutaneous tissues and 12.5% affected bone or joints (26). Osteomyelitis due to *P multocida* typically arises from direct inoculation of bone, or contiguous spread of superficial infection, and usually involves the distal extremities – the most common site of animal bites or scratches. In a review of 36 cases of osteomyelitis due to *P multocidal* two involved the axial skeleton and 34 involved distal extremities (26). Hematogenous spread from a distant site has been reported (27).

P multocida has been isolated from the human respiratory tracts both as commensal (25) and pathogen (25,28-31), usually in the presence of bronchiectasis or chronic bronchitis (29). Rare cases of *P multocida* sinusitis have been reported (25,29,30). In Weber et al's review (26), oral and respiratory infections were the second most common site of *P multocida* isolation in humans, accounting for 13.6% of infections.

Our patient was atypical in that she had only very limited, nontraumatic animal contact. Human infections due to *P multocida* have been reported after nontraumatic exposures (25,31) and in patients with no recognized exposures (25). In our patient, initial colonization of the respiratory tract was the most likely source of infection, with subsequent infection of an abnormal frontal sinus, followed by progression to osteomyelitis and PPT. Failure to recognize the clinical significance of frontal cellulitis led to delay in diagnosis and treatment, and failure to achieve early, definitive sinus drainage led to a delay in complete recovery.

SUMMARY

PPT represents a frontal subperiosteal abscess arising from osteomyelitis of the frontal bone. Children and young adults are most commonly affected, typically following sinusitis or trauma. When frontal cellulitis is present, underlying PPT should be considered even in the absence of systemic features of infection or prominent symptoms of sinusitis. Clinicians must be alert to the possibility of associated serious intracranial complications. Urgent CT or magnetic resonance image scanning of the cranium and sinuses is indicated to identify intracranial complications. Bone scintigraphy with ^{99}Tc-mMP may be more sensitive than CT scanning in the detection of early osteomyelitis (6) but lacks specificity in the context of acute sinusitis as a consequence of increased bone turnover in uninfected bone adjacent to areas of active infection. Early surgical intervention to establish long term sinus drainage is essential to avoid late complications. Initial empiric antibiotic therapy should provide broad spectrum activity with good central nervous system penetration. Surgical consultation for management of intracranial complications and sinus disease is essential.

ACKNOWLEDGEMENTS: The authors thank Ms Leslie Fisher for her help in preparing this manuscript and Drs S Valnicek, Division of Plastic Surgery, and B Tan, Division of Infectious Diseases, Royal University Hospital, Saskatoon, Saskatchewan for reviewing the manuscript.

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