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Unique presentation of *Aggregatibacter aphrophilus* in a superficial left temporal abscess

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ABSTRACT

Aggregatibacter aphrophilus, previously *Haemophilus aphrophilus*, is an uncommon organism that historically has been associated with HACEK infective endocarditis and brain abscesses. This organism is most often isolated as part of normal oral flora, and patients with *A. aphrophilus* infection usually have an underlying periodontal infection or immunocompromised state allowing for infection. This case report outlines a unique presentation of left superficial temporal abscess due to *A. aphrophilus* infection in an immunocompetent individual.

Introduction

Aggregatibacter aphrophilus, previously *Haemophilus aphrophilus*, is a gram-negative aerobic rod first described by Khairat in a case of endocarditis [1,2]. It has since emerged as an important, albeit rare, etiology of HACEK group (*Haemophilus, Aggregatibacter, Cardiobacterium, Eikenella, Kingella*) infectious endocarditis and brain abscesses [2]. This organism has low pathogenicity with slow growth resulting in its being an uncommon source of infection in immunocompetent patients [3]. *A. aphrophilus* is frequently found as part of normal oral flora and occurs in low proportions in subgingival plaque [4]. Previous cases of *A. aphrophilus* infection usually involve periodontal infection, previous dental procedure, or immunocompromise allowing opportunistic infection by this organism. In this text, we will present a case of *A. aphrophilus* causing a superficial left temporal abscess in a young, immunocompetent patient. This organism is an uncommon causative agent in abscesses and has not been previously implicated in superficial abscesses of the temporal region.

Case presentation

A 13-month-old female with no pertinent medical history was admitted with a one-week history of left temporal swelling. The patient's mother reported no previous trauma to the area and a lack of apparent tenderness on palpation. The mother also stated that the patient experienced a short viral illness approximately one to two months previous to presentation with apparent trismus. The patient had no residual symptoms of the viral illness and no longer appeared to demonstrate decreased range of motion of the jaw or decreased oral intake. The patient was up to date on all immunizations and had a normal birth and developmental history. All questions on review of systems were negative except for a temperature of 37.7 °C. On examination there was boggy, non-tender, non-erythematous swelling of the left temporal region. The patient was otherwise normocephalic with no other positive findings. Investigations revealed a white blood cell count of 17.6 K/uL (normal range 6–17.5 K/uL) with neutrophilia and C-reactive protein of 17.0 mg/dL (normal range 0.0−0.5 mg/dL).

CT of the maxillofacial region and sinuses with contrast showed an indeterminate process centered over the left temporalis muscle with considerations including vascular or lymphatic malformation, myositis, or injury (Fig. 1). Magnetic resonance angiography and venography of the brain without contrast showed normal venous and arterial anatomy. MRI of the brain with and without contrast showed the enhancing mass overlying the left temporalis muscle with no cranial involvement and was otherwise normal. Infection was suspected from the clinical picture and the patient was discharged with a one-week course of amoxicillin-clavulanic acid and scheduled for follow-up with otolaryngology.

On follow-up one week later, the patient had finished the antimicrobial with no reduction in temporal swelling. The mother reported no further complications or developments with the exception of a mild cough and rhinorrhea. The patient was scheduled for ultrasound-guided fine needle aspiration of the mass in order to further evaluate.

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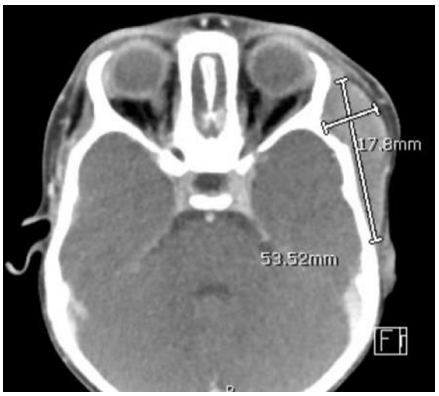

Fig. 1. CT of the maxillofacial region and sinuses with contrast showing a 17.8 mm x 53.52 mm mass over the left temporalis muscle.

However, the patient was readmitted six days after being seen in the clinic due to further enlargement of the mass. The mother stated that the swelling had nearly tripled in size, had become tender to touch, and the patient had decreased jaw opening. Repeat laboratory investigation showed a normal white count, but elevated erythrocyte sedimentation rate of 84 mm/hr (normal range 0–20 mm/hr) and C-reactive protein of 2.2 mg/dL (normal range 0.0–0.5 mg/dL). Repeat imaging with CT of the head with contrast showed enhancing soft tissue masses in the left frontal scalp, left masticator space, and left parotid space. MRI of the face and sinuses with and without contrast showed an infiltrative lesion in the left temporalis region with extension along the lateral aspect of left orbit and inferiorly into the infratemporal fossa. Differential considerations included cranial fasciitis or neoplastic processes such as rhabdomyosarcoma or fibrosarcoma. Infection was considered unlikely due to the stability in size compared to the previous admission's imaging (Fig. 2).

On day 2 of admission, excisional biopsy with incision and drainage of the left temporal region was performed. A 1 cm incision was marked over the most edematous region hidden in the hairline. Sharp incision was created through the skin down to temporal muscle. A wedge of temporalis muscle was removed and sent for histopathological evaluation. Blunt dissection was then taken down to the calvarium and down to the lateral orbital rim. Keratin debris with hair follicles were encountered and sent for histopathological evaluation. Cavity walls were also biopsied and sent for histopathological evaluation. The wound cavity was then copiously irrigated and suctioned and closed in layers.

All aerobic, anaerobic, and fungal cultures showed no growth. Histopathologic examination of the temporalis muscle and debris showed granulation tissue with nonspecific acute inflammation, lymphocytes, and macrophages. The patient was discharged with clindamycin 30 mg PO three times daily for seven days and scheduled for follow-up in otolaryngology clinic again.

During the next two clinic visits for follow-up, the course of antimicrobials were completed and the mother reported reduction in swelling and improvement in jaw opening. However, approximately one month later the patient presented to a follow-up visit with recurrence of left temporal swelling, this time with erythema and edema of the area. A seven-day course of amoxicillin-clavulanic acid was started and CT of the maxillofacial region with contrast and 3D reconstruction was performed in preparation for surgical removal. The imaging showed a stable size of the infiltrative lesion in the left temporalis muscle extending into the infratemporal fossa with underlying bony hyperostosis. There were enlarged lymph nodes in the left anterior parotid region. Differential considerations again included cranial fasciitis versus a neoplastic process (Fig. 3).

The patient was admitted one month later for excision of the left temporoparietal lesion. A 4 cm extended incision was performed at the site of the previous incision. At this point, sharp dissection was carried through the skin, subcutaneous tissue, and loose connective tissue until encountering the temporalis muscle. The muscle was noted to be very inflammatory with exudative material and some purulence. Samples of the exudate were sent for culture. Exudative tissue overlying the temporalis muscle was curetted off of the muscle and skin. This dissection was carried medially to the point of the epicenter of the lesion. There was noted to be a tract extending from the deeper tissue to the overlying skin. The tract was followed and removed. All tissue was sent for permanent pathology. Due to a small amount of bleeding, a TLS drain was placed during the postoperative period.

Pathology of the left temporal lesion showed granulation tissue with nonspecific acute and chronic inflammation. Cultures of the region returned showing 3+ beta lactamase negative *Aggregatibacter aphrophilus* colonies. Sensitivities of the organism were not available, so empiric intravenous azithromycin 94 mg once daily was begun. Bartonellosis and coccidioidomycosis serologies as well as a tuberculosis interferon- □ release assay were done at this time. The patient was discharged on day three of admission after

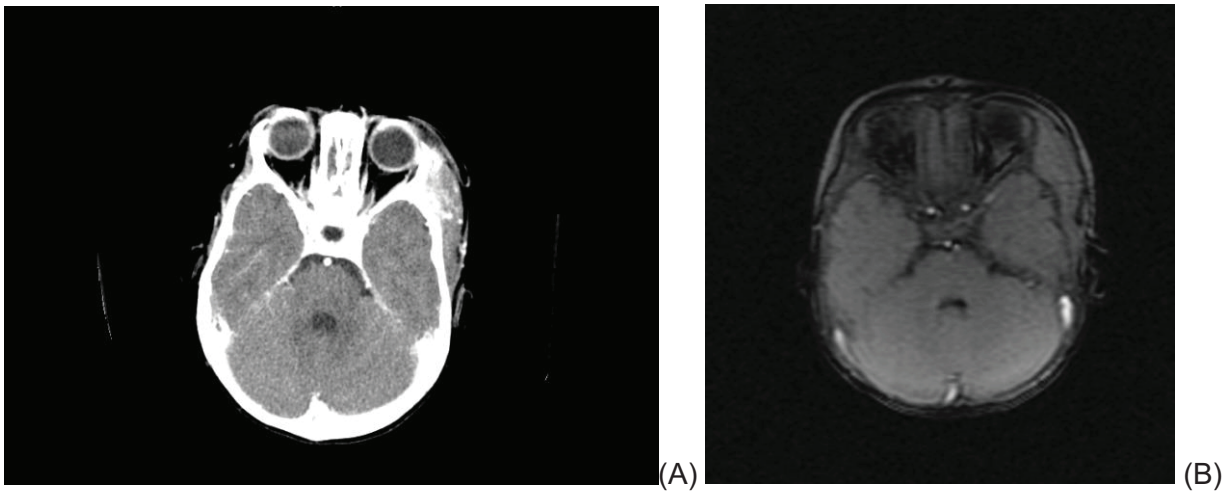

Fig. 2. (A) CT of the head with contrast showing enhancing soft tissue masses in the left frontal scalp, left masticator space, and left parotid space. (B) MRI of the face and sinuses showing an infiltrative lesion in the left temporalis region with extension along the lateral aspect of the left orbit and inferiorly into the infratemporal fossa.

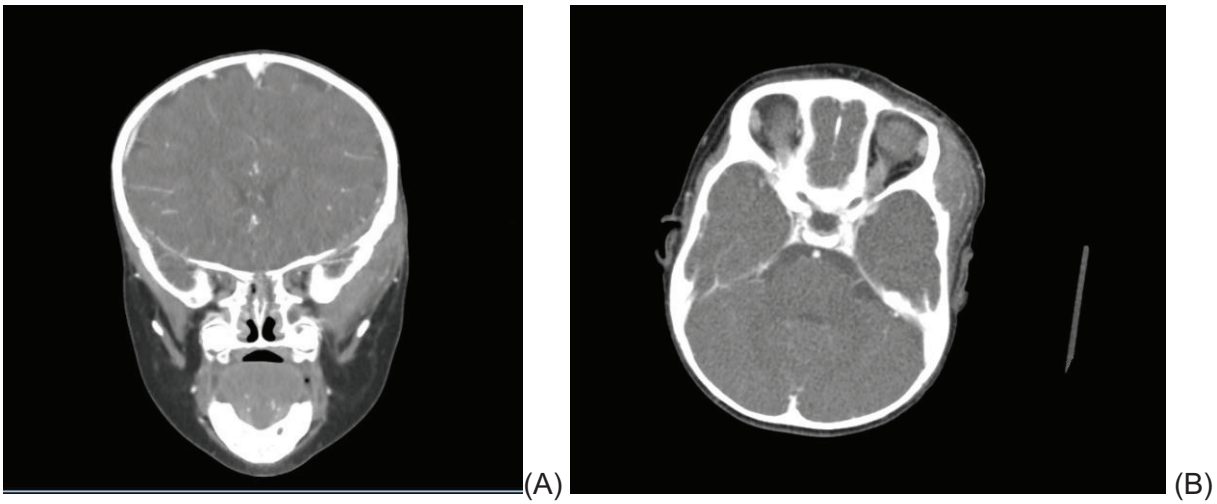

Fig. 3. (A) Frontal and (B) horizontal views of the CT of the maxillofacial region with contrast showing the infiltrative lesion in the left temporalis muscle extending into the infratemporal fossa with underlying bony hyperostosis.

removal of the TLS drain and transitioned to oral azithromycin 100 mg daily for seven days and cefixime 100 mg daily for ten days.

Upon follow-up with both pediatric infectious disease and otolaryngology, the patient had improved with no complications. The incision healed appropriately, antimicrobials were finished and discontinued, and bartonella, coccidioidomycosis, and tuberculosis testing was negative. The most recent laboratory investigations approximately one month after the final hospital admission showed a white count of 7.3 K/uL (normal range 6–17.5 K/uL), erythrocyte sedimentation rate of 13 mm/hr (normal range 0−20 mm/hr), and C-reactive protein of 0.1 mg/dL (normal range 0.0−0.5 mg/dL). The mother stated that swelling, trismus, fevers, and other symptoms have not recurred.

Discussion

The described case is the first reported of *A. aphrophilus* as the causative agent in a superficial abscess of the temporal region. This is also one of few previously reported cases of this organism in abscesses of the superficial head and neck region. Most previous reports of *A. aphrophilus* infections of this region involve canaliculitis [5,6] or exogenous endophthalmitis [7].

Initial imaging studies showed a mass overlying the left temporalis muscle, while later imaging studies showed infiltration extending to the infratemporal fossa to the point of questionable involvement of underlying bone. During surgical excision the extent of infiltration was confirmed as the infection was found to have penetrated fascia causing exudative debris and a small fistula from underlying tissue to the external skin. This is also distinct from previous presentations of abscesses caused by *A. aphrophilus;* most infections caused a well-encapsulated pocket of purulence that is classic for most abscesses of any origin. This suggests that this organism has the potential to cause deeper invasion, cosmetic deformity, and morbidity in immunocompetent patients. The delay in diagnosis highlights the importance of early suspicion and antibacterial coverage for *A. aphrophilus* in abscesses of the head and neck region.

Furthermore, extensive imaging with both CT and MRI could not differentiate between abscess formation and solid neoplastic process. In fact, abscess formation was considered to be unlikely due to the stable size of the lesion over time. The chronic infection with nonspecific clinical findings in this case also contributed to making radiologic diagnosis difficult. This difficulty has been encountered in the past with atypical infections of the infratemporal fossa and evaluation with histopathology and culture has proven to be more important for accurate diagnosis than radiologic findings in these cases [8].

Abscesses of the head and neck region are most commonly caused by oral flora [9]. Common anaerobes include *Prevotella, Porphyromonas, Fusobacterium,* and *Peptostreptococcus* species. The most commonly isolated aerobes are *Streptococcus pyogenes* and *Staphylococcus aureus,* which frequently cause skin and soft tissue abscesses anywhere on the body including the head and neck region. *A. aphrophilus* is most commonly isolated from normal oral flora which points to subacute odontogenic infection as the source in this case. The subsequent finding of extension into the infratemporal fossa also suggests a high likelihood of contiguous infection. There was no evidence of concurrent periodontal disease, underlying immunocompromise, or localized tissue damage in this patient at the time of presentation, but the short viral illness preceding the abscess described by the mother that was associated with trismus may be evidence of some sort of periodontal infection that caused contiguous spread. Although the source of infection cannot be definitively understood in this case, it nevertheless highlights the importance of keeping organisms native to oral flora in the differential for seemingly superficial abscesses of the head and neck region.

Initial empiric coverage was initiated with two courses of amoxicillin-clavulanic acid and one course of clindamycin, which were neither able to eliminate this patient's infectious process. The absence of growth of *A. aphrophilus* or any other organisms in this patient's initial cultures suggest two separate hypotheses: either cultures were not sufficiently taken in the procedure or *A. aphrophilus* was not initially present and the abscess had a different etiology that was further complicated by *A. aphrophilus* infection.

A. aphrophilus is most commonly sensitive to nearly all antimicrobial therapy including β-lactamase inhibitor combinations, third generation cephalosporins, meropenem, fluoroquinolones, and rifampin [10]. The normally broad antimicrobial sensitivity of this organism as well as the absence of growth on initial cultures point to the latter of the two formerly mentioned hypotheses. Our belief is that this patient most likely had a subacute periodontal infection, unrecognized trauma to the area, or other inciting event that initiated the abscess and was followed by opportunistic *A. aphrophilus* colonization.

After diagnosis of *A. aphrophilus* on the second set of cultures, coverage with azithromycin and cefixime along with surgical

debridement resulted in complete recovery. Broad-spectrum cephalosporins and fluoroquinolones are most commonly used for antimicrobial treatment of HACEK organisms based on past reports and proved to eliminate this patient's infection and support the use of these antimicrobials for future cases [11].

Conclusion

In this report, we described a case of superficial left temporal abscess caused by *A. aphrophilus* without clear evidence of contiguous odontogenic infection or underlying immunocompromise. This is the first known reported case of *A. aphrophilus* causing a superficial abscess in the temporal region and suggests that this organism has the ability to cause other infections of the head and neck region besides the previously described ophthalmic disease. The recurrent abscess and delay in diagnosis in this case highlights the need for a high index of suspicion for unusual organisms in unexplained abscesses.

CRediT authorship contribution statement

Tanir Moreno: Writing - original draft. **Rahul Varman:** Writing - review & editing, Supervision. **Winslo Idicula:** Writing - review & editing, Supervision.

Declaration of Competing Interest

None.

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