#### **CASE REPORT**

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# Actinomyces turicensis parapharyngeal space infection in an immunocompetent host: first case report and review of literature

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#### **Abstract**

Actinomyces are common commensals of the oral cavity, gastrointestinal tract and urogenital tract. They are anaerobic, Grampositive, non-acid-fast bacilli, which can cause invasive infection and abscesses. We present the first reported case of supraglottitis and deep neck space abscess formation secondary to Actinomyces turicensis infection. The patient was managed with intravenous antibiotics, incision and drainage of a left parapharyngeal abscess and subsequent mediastinal abscess. After 6 weeks in hospital, the patient was successfully discharged to complete a 6-month course of oral amoxicillin.

## INTRODUCTION

Actinomycosis is a granulomatous condition characterized by subacute or chronic abscess formation. The responsible Actinomyces organisms are anaerobic, Gram-positive, nonspore-forming, non-acid-fast, filamentous, branching rods that are normal commensals of the oral cavity, gastrointestinal tract and urogenital tract. Severe infections can result if they breach the submucosa. Actinomycosis infection has been divided into three clinical forms: cervicofacial (50%), thoracopulmonary (30%) and abdominopelvic (20%). Actinomyces israelii is the most common causative agent in cervicofacial actinomycosis. Conversely, Actinomyces turicensis is rare [1]. Predisposing factors for cervicofacial infection include recent dental procedures, immunosuppression, smoking and excessive consumption of alcohol.

Supraglottitis is an infection of the epiglottis and surrounding structures, including the arytenoids, false vocal cords and laryngeal ventricles. It is a life-threatening condition due to the potential for upper airway obstruction, and is more common in males, with a similar risk factor profile to actinomycosis infections [2]. To our knowledge, this is the first reported case of A. turicensis implicated in deep space neck abscess as a complication of supraglottitis.

# **CASE REPORT**

A 56-year-old man presented to the emergency department with a 3-day history of sore throat and worsening left-sided neck fullness, associated with odynophagia, dysphagia and fever. He reported a fit and active lifestyle with a history of well-controlled asthma. No risk factors for deep neck space infection were elicited from his medical background.

Upon assessment by the otolaryngology team, inspiratory stridor was noted. Flexible nasolaryngoscopy revealed epiglottic oedema and swelling in the left vallecula. His white cell count was 13.1×109 l-1 and his C-reactive protein value was 306 mg/l. He was started on intravenous dexamethasone, ceftriaxone and metronidazole for empirical treatment of supraglottitis. Repeat flexible nasolaryngoscopy demonstrated progressive supraglottic oedema and impending airway obstruction. He was jointly managed with the anaesthetic team, intubated in theatre and transferred to the intensive therapy unit (ITU).

Computed tomography (CT) of the neck with contrast revealed extensive supraglottitis with left-sided phlegmon formation, but no evidence of a drainable abscess (Fig. 1). Inflammatory changes were seen within the superior mediastinum that were suggestive of superior mediastinitis. Intravenous ceftriaxone and metronidazole were continued.

Following a 3-day interval, due to minimal clinical improvement, a repeat CT scan was performed (Fig. 2). This revealed disease progression with probable abscesses tracking anteriorly and laterally to the thyroid cartilage. Incision and drainage were performed and pus was evacuated from the left parapharyngeal and central suprahyoid spaces. Tissue

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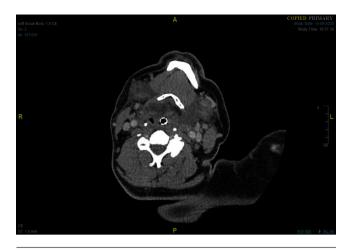
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Keywords: Actinomyces; actinomycosis; parapharyngeal space; supraglottitis.

Abbreviation: NSTI, necrotising soft tissue infection.

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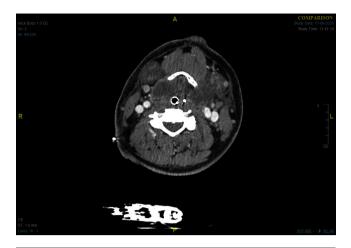


**Fig. 1.** Computed tomography (CT) of the neck with contrast, axial plane, performed on the day of presentation, revealing extensive supraglottic inflammatory changes but no sign of drainable abscess.

submitted for histological analysis showed fibro fatty tissue with foci of acute inflammation, without atypia or malignancy.

The evacuated pus sent to the microbiology laboratory was inoculated onto blood agar plates incubated in added CO. at 35-37 °C, a MacConkey agar incubated aerobically at 35-37 °C and a chocolate agar plate with a metronidazole 5 μg disc incubated anaerobically. In addition, enrichment cultures in Robertson cooked meat broth were incubated at 35–37 °C. Cultures of the abscess revealed moderate growth of A. turicensis sensitive to penicillin; Enterococcus faecalis sensitive to amoxicillin, teicoplanin, vancomycin and linezolid; Streptococcus anginosus sensitive to penicillin, clindamycin and erythromycin; and moderate growth of Streptococus constellatus sensitive to penicillin, levofloxacin and vancomycin and resistant to erythromycin, clindamycin and tetracycline. All the organisms, including A. turicensis, were identified using matrix-assisted desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS) and antimicrobial susceptibility was determined using the minimum inhibitory concentration (MIC) per the European Committee on Antimicrobial Susceptibility Testing (EUCAST) methodology.

Four days post-operatively, the patient became persistently pyrexial and blood tests revealed a raised white cell count. The antibiotics were changed to meropenem and metronidazole based on microbiological advice. A repeat CT scan was performed which noted mediastinal abscess formation (Fig. 3.). Electrocardiography (ECG) revealed widespread ST elevation, compared to a baseline normal sinus rhythm. Echocardiography demonstrated evidence of a pericardial effusion. The troponin T was not elevated, and these changes were diagnosed as pericarditis. The patient underwent an incision and drainage of mediastinal abscess and surgical tracheostomy, jointly performed by the otolaryngological and cardiothoracic teams. Copious purulent liquid was expressed from the superior and posterior mediastinum. Cultures



**Fig. 2.** CT of the neck with contrast, axial plane, performed 3 days following initial presentation, revealing disease progression and small abscess/phlegmon noted anterolateral to both sides of thyroid cartilages.

here revealed *E. faecalis* sensitive to amoxicillin, linezolid, vancomycin and teicoplanin with extended-spectrum betalactamase-producing *Escherichia coli* (both on enrichment only) and mixed anaerobes.

Meropenem and metronidazole were stopped after 2 weeks. In light of the *Actinomyces* isolate, intravenous amoxicillin 2 g three times daily was commenced for 3 weeks, with a plan to switch to oral amoxicillin 1 g three times daily for 6 months.

Six weeks after admission, the patient was decannulated, able to mobilize with a frame and was recommencing oral nutrition. He was successfully discharged home.

At the time of writing (2 months following discharge from hospital), the main sequela reported by the patient is a higher pitched voice, although this is gradually improving. He is managing oral intake at a pre-morbid level and has

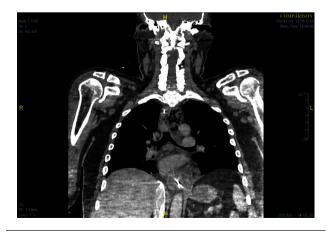


Fig. 3. CT of the neck and thorax with contrast, coronal plane, performed 7 days following initial presentation, revealing a mediastinal collection measuring  $36\times34$  mm above the level of the aortic arch.

Continued

Table 1. Reported cases of Actinomyces turicensis infection

Reference	Age	Diagnosis	Concomitant organisms	Risk factor(s)	Country	Treatment	Sequelae
Riegert-Johnson <i>et al.</i> , 2002 [5]	59	Hepatic abscess	Bacteroides fragilis	Recent dental procedure	USA	Antibiotics (5 weeks)	None
Attar et al., 2007 [6]	33	Right breast abscess	Mixed anaerobes	Obesity Ulcerative colitis	UK	Antibiotics (3 weeks) Surgical	None
Zautner <i>et al.</i> , 2009 [7]	23	Right knee joint	Actinomyces europaeus	Fistulae excision 6 months prior	Germany	Antibiotics (2 weeks) Surgical	None
Chudácková <i>et al.</i> , 2010 [8]	18	Pilonidal cyst	None	None known	Czech Republic	Surgical+/-antibiotics	None
Chudácková <i>et al.</i> , 2010 [8]	18	Anorectal abscess	Bacteroides ureolyticus, Fusobacterium nucleatum	None known	Czech Republic	Surgical+/-antibiotics	None
Chudácková et al., 2010 [8]	28	Buttock abscess	None	None known	Czech Republic	Surgical+/-antibiotics	None
Chudácková <i>et al.</i> , 2010 [8]	23	Perianal abscess	Streptococcus milleri, Peptostreptococcus anaerobius	None known	Czech Republic	Surgical+/-antibiotics	None
Chudácková et al., 2010 [8]	28	Pilonidal abscess	Staphylococcus aureus	None known	Czech Republic	Surgical+/-antibiotics	None
Chudácková et al., 2010 [8]	33	Buttock abscess	Propionibacterium acnes	Diabetes mellitus	Czech Republic	Surgical+/-antibiotics	None
Chudácková <i>et al.</i> , 2010 [8]	65	Scrotal gas gangrene	Prevotella spp.	Obesity Rheumatoid arthritis Diabetes mellitus Hypertension	Czech Republic	Antibiotics Surgical	None
Ong, Barnes and Senanayake, 2012 [9]	73	Left iliac fossa collection	None	Obesity	Australia	Antibiotics (30 weeks)	None
Miller <i>et al.</i> , 2014 [10]	25	Right cerebellar abscess secondary to otitis media	Proteus mirabilis, Peptoniphilus harei, Bacteroides thetaiotaomicron, Anaerococcus hydrogenalis	None known	New Zealand	Antibiotics (>5 weeks) Surgical	Right lateral gaze palsy
Abdulrahman and Gateley, 2015 [11]	22	Right breast abscess	P. harei, Staphylococcus epidermidis	Nipple piercing >1 year prior	UK	Antibiotics (>26 weeks) Needle aspiration	None
Kottam et al., 2015 [12]	30	Eustachian valve endocarditis and hepatic abscess	None	Intrauterine device placed 2 years ago	USA	Antibiotics (8 weeks) Surgical	None
Hagiya <i>et al.</i> , 2015 [13]	80	Pyometra	Clostridium clostridioforme, Escherichia coli	None known	Japan	Antibiotics (4 weeks) Transvaginal drainage	None
Oh, Abdul Malik and Keh, 2015 [14]	25	Pilonidal abscess	Prevotella bivia, Peptostreptococcus spp.	None known	Singapore	Antibiotics (1 week) Surgical	None
Eenhuis et al., 2016 [15]	42	Pelvic-abdominal peritonitis	Anaerobes	Intrauterine device placed 5 years ago	The Netherlands	Antibiotics (30 weeks) Surgical	None
Gatti et al., 2017 [16]	64	Anterior abdominal wall NSTI	None	Obesity Hypertension	Italy	Antibiotics (5 weeks) Surgical	None
Cobo, 2018 [17]	44	Right breast abscess	None	None known	Spain	Antibiotics (10 days)	None
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None

Antibiotics (7 weeks)

UK

Sequelae Death

Treatment

None

Antibiotics Surgical

USA

Antibiotics Surgical

Country Hungary None

Antibiotics (6 weeks) Surgical

USA

Not known

Antibiotics (11 weeks)
Percutaneous drainage
Antibiotics (26 weeks)
Surgical

PR China

USA

None

None

Antibiotics (32 weeks) Surgical

UK

None

Antibiotics (2 weeks)

USA

Surgical

Previous radiotherapy for Obesity Diabetes mellitus Persistent lactation Recent oral surgery Poor oral hygiene Ureteric stones Risk factor(s) Alcohol abuse None known None known oral cancer Enterococcus faecalis, mixed anaerobes, Streptococcus anginosus, Streptococcus constellatus E. coli, P. mirabilis, anaerobes Concomitant organisms Peptostreptococcus spp. Anaerobes P. harei None None None Prostate, left inguinal cord and right facial abscess Supraglottitis, parapharyngeal and mediastinal abscess Cervicofacial actinomycosis Mastoiditis and meningitis Pyelonephritis and abscess Adrenal gland abscess Right breast abscess Right thigh NSTI Diagnosis Age 43 45 61 43 52 20 53 26 Kansara et al., 2020 [22] Panwar et al., 2019 [19] Barnes, Kaur and Augenbraun, 2020 [24] Table 1. Continued Kocsis et al., 2018 [18] Le Bihan, Ahmed and Vassa et al., 2019 [20] O'Driscoll, 2019 [21] Jin et al., 2020 [23] Current case Reference

not suffered any recurrent or new infections. Screening for an underlying immunodeficiency has been negative and he remains under regular review.

## DISCUSSION

This is the first case of A. turicensis reported as a causative organism in deep neck space infection arising from supraglottitis. Although a mucosal commensal and found within a polymicrobial abscess, it is extremely rare, and clinically significant when isolated from sterile deep-seated locations [1]. Notably, while deep neck space and cervicofacial Actinomyces infections have risk factors in common, the patient featured in this case had none of these reported in their history, and further investigations have not yielded an underlying cause. Growth of Actinomyces can take up to 15-20 days and due to its facultatively anaerobic character, careful transport and anaerobic processing of specimens suspected to harbour Actinomyces are required, which may partially account for its rarity. Penicillin G and amoxicillin are the antibiotics of choice for Actinomyces infections because of their susceptibility to beta-lactams. Patients typically require a prolonged course of treatment, for a maximum of 12 months, with a minimum of 3 months where optimal surgical resection is thought to have been achieved [3].

Since the advent of widespread immunization against Haemophilus influenzae, supraglottitis is most commonly caused by Streptococcus pneumoniae, Staphylococcus aureus and Neisseria meningitidis. Non-bacterial causes, including viruses, trauma, chemoradiotherapy and chemical irritants, are less common [2]. Some cases may mimic malignancy or cancer relapse [3]. Adults with supraglottitis commonly present with severe sore throat, odynophagia, dysphagia, dysphonia, pyrexia and, less commonly, stridor. There may be associated cervical lymphadenopathy and anterior neck tenderness, but not necessarily in the early stages. Diagnosis is made by flexible nasolaryngoscopy to reveal erythema and oedema of the supraglottis. Acute management includes supplemental oxygen, nebulized adrenaline, intravenous dexamethasone and empirical intravenous antibiotics in line with local antimicrobial guidelines. Tracheostomy or endotracheal intubation are required in patients with severe oedema and those who are unresponsive to medical management. There is sparse evidence in the literature quantifying supraglottitis progression to abscess formation, but one multicentre study involving 202 patients suggested a 22% incidence of epiglottic abscess in patients with acute supraglottitis [2, 4].

Isolates of *A. turicensis* have most commonly been described to involve the abdomen and soft tissue outside the head and neck. There are 26 previously reported cases of *A. turicensis* causing infection in humans, but no reports of association with supraglottitis or deep neck space abscess. Table 1 summarizes a literature review of *A. turicensis* infections. The 26 cases include 9 with *A. turicensis* only, and 17 with concomitant organisms. A predisposing factor was reported in 16 cases. At least 22 cases included abscess formation requiring 1 or more surgical procedures, which is replicated

in our report. Notably, low mortality has been recorded, as all but one case survived. It is unclear why the patient in our case had *A. turicensis* isolated in the parapharyngeal space – on questioning he had no reported abdominal or genitourinary symptoms prior to his presentation.

A. turicensis can feature as a causative micro-organism in supraglottitis, and may predispose to higher disease severity in the context of abscess formation within the deep neck spaces. Where A. turicensis is isolated, clinicians must be prepared to intervene surgically as part of the management plan. Microbiological samples should be obtained and transported carefully and promptly, as A. turicensis is a facultative anaerobe. This is key to determining the correct antimicrobial treatment. In our case, the first attempts to treat the infection (with ceftriaxone and metronidazole) were unsuccessful. A long duration (at least 6 months) of amoxicillin or penicillin G is indicated, and a multidisciplinary approach is essential for successful patient management.

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#### Conflicts of interest

The authors declare that there are no conflicts of interest.

#### Ethical statement

Verbal and written consent was obtained from the patient.

#### References

- Valour F, Sénéchal A, Dupieux C, Karsenty J, Lustig S, et al. Actinomycosis: Etiology, clinical features, diagnosis, treatment, and management. Infect Drug Resist 2014;7:183–197.
- 2. Navaratnam AV, Smith ME, Majeed A, McFerran DJ. Adult supraglottitis: A potential airway emergency that can present in primary care. *Br J Gen Pract* 2015;65:99–100.
- Choudhury S, Agrawal A, Rane S, Rangarajan V. Serendipitous actinomycosis infection mimicking malignant lesion in carcinoma of unknown primary. World J Nucl Med 2019;18:413–415.
- Lee YC, Kim TH, Eun YG. Routine computerised tomography in patients with acute supraglottitis for the diagnosis of epiglottic abscess: Is it necessary?--a prospective, multicentre study. Clin Otolaryngol 2013;38:142-147.
- Riegert-Johnson DL, Sandhu N, Rajkumar SV, Patel R. Thrombotic thrombocytopenic purpura associated with a hepatic abscess due to Actinomyces turicensis. Clin Infect Dis 2002;35:636–637.
- Attar KH, Waghorn D, Lyons M, Cunnick G. Rare species of Actinomyces as causative pathogens in breast abscess. Breast J 2007;13:501–505.
- Zautner AE, Schmitz S, Aepinus C, Schmialek A, Podbielski A, et al. Subcutaneous fistulae in a patient with femoral hypoplasia due to Actinomyces europaeus and actinomyces turicensis. Infection 2009;37:289–291.
- 8. Chudácková E, Geigerová L, Hrabák J, Bergerová T, Liska V, et al. Seven isolates of Actinomyces turicensis from patients with surgical infections of the anogenital area in a czech hospital. J Clin Microbiol 2010;48:2660–2661.
- 9. Ong C, Barnes S, Senanayake S. *Actinomyces turicensis* infection mimicking ovarian tumour. *Singapore Med J* 2012;53:e9–e11.
- Miller S, Walls T, Atkinson N, Zaleta S. A case of otitis media complicated by intracranial infection with Actinomyces turicensis. JMM Case Rep. 2014;1:e004408.
- 11. **Abdulrahman GO, Gateley CA**. Primary actinomycosis of the breast caused by *Actinomyces turicensis* with associated *Peptoniphilus harei*. *Breast Dis* 2015;35:45–47.

- 12. Kottam A, Kaur R, Bhandare D, Zmily H, Bheemreddy S, et al. Actinomycotic endocarditis of the eustachian valve: A rare case and a review of the literature. Tex Heart Inst J 2015;42:44–49.
- 13. **Hagiya H**. *Actinomyces Turicensis* bacteremia secondary to pyometra. *Intern Med Open Access* 2015;54:2775–2777.
- Oh HB, Abdul Malik MH, Keh CHL. Pilonidal abscess associated with primary actinomycosis. Ann Coloproctol 2015;31:243–245.
- 15. **Eenhuis LL**, **de Lange ME**, **Samson AD**, **Busch ORC**. Spontaneous bacterial peritonitis due to actinomyces mimicking a perforation of the proximal jejunum. *Am J Case Rep* 2016;17:616–620.
- Gatti M, Gasparini LE, Grimaldi CM, Abbati D, Clemente S, et al. Septic shock due to NSTI caused by Actinomyces turicensis: the role of clinical pharmacology. Case report and review of the literature. J Chemother 2017;29:372–375.
- Cobo F. Breast abscess due to Actinomyces turicensis in a nonpuerperal woman. Enferm Infecc Microbiol Clin 2018;36:388–389.
- Kocsis B, Tiszlavicz Z, Jakab G, Brassay R, Orbán M, et al. Case report of Actinomyces turicensis meningitis as a complication of purulent mastoiditis. BMC Infect Dis 2018;18:686.

- 19. Panwar K, Duane TM, Tessier JM, Patel K, Sanders JM, et al. Actinomyces turicensis necrotizing soft-tissue infection of the thigh in a diabetic male. Surg Infect (Larchmt) 2019;20:431–433.
- 20. Vassa N, Mubarik A, Patel D, Muddassir S. *Actinomyces turicensis*: An unusual cause of cervicofacial actinomycosis presenting as ludwig angina in an immunocompromised host Case report and literature review. *IDCases* 2019;18:e00636.
- 21. Le Bihan A, Ahmed F, O'Driscoll J. An uncommon cause for a breast abscess: with. *BMJ Case Rep* 2019;12:12.
- 22. Kansara T, Majmundar M, Doshi R, Ghosh K, Saeed M, et al. A case of life-threatening Actinomyces turicensis bacteremia. Cureus 2020:12:e6761.
- 23. Jin W, Miao Q, Wang M, Zhang Y, Ma Y, et al. A rare case of adrenal gland abscess due to anaerobes detected by metagenomic next-generation sequencing. Ann Transl Med 2020;8:247.
- Barnes A, Kaur A, Augenbraun M. An unusual presentation of prostatic abscess due to actinomyces turicensis and *Peptostrepto*coccus. Cureus 2020;12:e8665.

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