

Metastatic Gas Gangrene of the Neck Due to Local Trauma to the Upper Extremity, Case Report and Literature Review



Abdulaziz AlEnazi^{1*}, Dinah AlNoaimi², Mohammed AlWabili³ and Aliya AlMutairi³

¹Department of Otolaryngology- Head and Neck Surgery, Al-Khobar, Imam Abdul Rahman bin Faisal University, Saudi Arabia

²Department Otolaryngology-Head and Neck Surgery, King Fahad Specialist Hospital, Saudi Arabia

³Department of Otorhinolaryngology-Head and Neck Surgery, Prince Sultan Military Medical City, Saudi Arabia

Submission: October 22, 2022; **Published:** October 28, 2022

***Corresponding author:** Abdulaziz Saud AlEnazi, Department of Otolaryngology-Head and Neck Surgery, College of Medicine, King Fahd Hospital of the University, Al-Khobar, Imam Abdul Rahman bin Faisal University, Dammam, Saudi Arabia

Abstract

Background: Metastatic, non-traumatic, or spontaneous gas gangrene is a rare fulminant clinical entity associated with rapid evolution of clinical signs and symptoms. It is highly linked to clostridial species and *Clostridium perfringens* is the most common bacterium causing gas gangrene. The exact mechanism behind it is not fully understood yet. moreover, high mortality rates are seen with it and are related to host factors, healthcare factors, and anatomical location of the infection. However, if recognized early and managed properly the mortality rate drop to approximately 20 to 30%.

Case presentation: We report a case of a healthy young 18-year-old female who is a known case of metastatic gas gangrene since childhood after sustaining a left-hand fracture. She presented to the emergency department of prince sultan military medical city in Riyadh, Saudi Arabia with 2 days history of neck pain and crepitation over the left side of her neck extending superiorly up to the left cheek on examination with no signs of infection or fever. Our patient was managed conservatively after thorough investigations and computed tomography with contrast of the neck.

Conclusion: We consider our case to be the first in the English literature to be reported in this area carrying additional significance of developing such lethal condition in an extremely viable structure like the neck.

Keywords: Gas gangrene; Metastatic gas gangrene; Trauma; Infection; Neck pain

Introduction

Metastatic, non-traumatic, or spontaneous gas gangrene, a term which has been gaining attention lately is used to describe gas gangrene if develop without any antecedent trauma [1] and it is largely linked to clostridium species namely *C. septicum* (clostridium septicum) [2]. Although the condition is rare, it carries a high mortality risk and the development of clinical picture is extremely rapid [3]. Many attempts have been carried out to understand the exact factors that provoke the development of this condition, however, the exact mechanism behind such condition is still not fully understood [1].

Moreover, metastatic gas gangrene has been largely known to be associated with serious medical conditions such as gastroin-

testinal and hematological malignancies, immunocompromising diseases like diabetes mellitus, or peripheral vascular diseases [4]. Also, iatrogenic interventions like colonoscopy, radiation, or surgery might increase the risk of this condition. I Srivastava et al. [1] recent review of the literature has identified 94 cases of spontaneous gas gangrene from 1956 to 2016, they analyzed each case by characterizing its clinical picture, the focus of infection, and therapeutic interventions, however, no single case of metastatic gas gangrene to the neck has been reported. We consider our case to be the first in the English literature to be reported in this area carrying additional significance of developing such lethal condition in an extremely viable structure like the neck.

Case Presentation

This is a case of an 18-year-old female who's a known case of metastatic gas gangrene since she was 4-year-old. At that age, the patient sustained a left-hand fracture which was reduced surgically by the orthopedic team. Unfortunately, a while after, the patient developed a surgical site infection which was too rapid leading to gas gangrene that worsened to the point of involving her whole body. After appropriate management with systemic antibiotics, proper analgesia, and hyperbaric oxygen, the patient improved and was able to go home. Recently, our patient presented to the Emergency room with a complaint of sudden neck pain of two days duration. The pain was intermittent, yet it wasn't associated with fever, hoarseness, shortness of breath, or dysphagia. It was relieved by one dose of Intravenous morphine.

Upon examining the patient was conscious, alert, and oriented. Not in pain or distress having a poor body built. Vitals were taken she had a heart rate of 78 beat per minute, a blood pressure of 109/59mmhg and she was afebrile. During Neck Examination, there was no goiter, or any mass noted. However, on palpation, she had crepitation over the left side of her neck extending superiorly up to the left cheek, with no palpable lymph nodes. Her nose, throat, and ear examinations were unremarkable. Her blood investigation and chemistry were within normal limits apart from low hemoglobin of 9.1g/dL and a slightly elevated Erythrocyte sedimentation rate of 22mm/hr. No signs of infection were noted.

Our patient underwent Computed tomography of the neck with contrast enhancement to evaluate the finding in her neck. It revealed significant multiple large interconnecting air pockets identified in the facial region and the neck on both sides. They were specifically along the left side, anterior to the sternocleidomastoid muscle and within the muscle fascia. The air was dissecting within the planes of the muscles and reaching superiorly to the submandibular region with extension to the posterior neck triangle. The gas was also identified in her cheeks' region bilaterally and masseter muscles which were more pronounced on the left side. The air pockets in bilateral cheeks were associated with significant facial swelling and right-sided facial edema. Fortunately, no significant lymphadenopathy was noted.

Additionally, and after a thorough discussion and review of similar cases in the literature. Our patient underwent full body computed tomography to evaluate for any intraabdominal or intrathoracic pathology. Fortunately, all of her radiologic and Histopathological assessments were not significant. The decision was made to manage her conservatively.

Discussion

Metastatic Gas Gangrene

Spontaneous, non-traumatic, or metastatic gas gangrene are all different synonyms for the same clinical entity. Although not common, this condition has been largely known to be lethal. The

underlying pathophysiology is still not established; however, it's known to be caused by *C. septicum* species when they gain access to the bloodstream due to any mucosal pathology especially in the colon [5]. It is also reported that gas gangrene is highly associated with penetrating or surgical wounds, gastrointestinal surgeries, immunocompromising diseases, hematological or gastrointestinal malignancies, or traumatic injuries. However, it could arise spontaneously. Based on this information, our patient underwent a thorough assessment for any intrabdominal pathology including radiological and blood evaluations.

Interestingly, all of her tests showed no intrabdominal abnormalities. According to the last review done by I Srivastava et al. [1]. [most of the reported cases were associated with preexisting diabetes mellitus (41%), one explanation of this finding could be linked to the effect of diabetes on neutrophils function causing functional neutrophilia [6]. This effect can lead to weak mucosal functional immunity rendering the bacteria with more ability to travel through and out to distant locations. However, our patient didn't have hyperglycemia of any type and her immunology profile was within the normal level. Additionally, regarding the site of the infection, no single case in their review involved the neck which was a very critical finding in our case [7].

Causative Organism

Clostridial gas gangrene is caused by anaerobic, spore-forming clostridium subspecies that result in life-threatening consequences. As previously mentioned, it could arise in traumatized tissue or less commonly spontaneously. *C. septicum* is a gram-positive opportunistic pathogen in humans normally present in gastrointestinal normal flora, bacteria gain access into the circulation through breakings in the mucosal linings. This is usually brought upon by the presence of a tumor or necrotizing enterocolitis in a patient diagnosed with hematological malignancies or neutropenia. Assuming that a tumor provides an advantageous environment giving clostridial spores the opportunity to germinate [8,9]. As documented by Thiele et al. [10] the affinity of necrotic tissue is not exclusive for malignant tissue, Seeing that spores of Clostridia were injected and germinated in necrotic tissue other than that of a tumor. *C. septicum* might result in a contained local infection or migrate through the circulation to other organs and muscles resulting in systemic toxicity [10].

Mechanism

The exact mechanism of this condition is still unknown to this day. Many theories have been developed. The organism releases toxins that lead to the progressive nature and immediate spread of clostridial infection that may cause systemic toxicity. *C. septicum* produces four main toxins, alpha-toxin (hemolyzing necrotizing toxin). In addition, it also produces beta-toxin (deoxyribonuclease), gamma-toxin (hyaluronidase), and the delta-toxin (hemolysin toxin). creatinine phosphokinase which is a compounded released upon tissue degradation could result in secondary toxicity. In a case report it was seen that diabetics are susceptible to devel-

oping gas gangrene spontaneously in 41% of the cases [11]. This is given to their tendency to develop tissue ischemia and acidosis as a result of ongoing atherosclerosis in addition to, microangiopathic vascular disease, which makes it favorable to the organism to spread in areas that are hypoxic [4]. *C. septicum* gas gangrene was also found to be associated with neutropenia, immunosuppression, and malignancy, mainly hematological and gastrointestinal malignancies.

Clinical Presentation

Gas gangrene is a rare entity that many surgeons are unfamiliar with its clinical picture and do not immediately recognize its presence and progression. The major hurdle in the diagnosis is rooted in the struggle of differentiating a true infection from simple contamination. In addition to that, the nonspecific nature of gas gangrene signs and symptoms and the other types clostridial infections and other lesions similar to gas gangrene makes it difficult to reach an accurate diagnosis in a short duration of time. The finding of gram-positive anaerobic bacilli in a necrotic wound should raise the suspicion of the presence of gas gangrene [12,13]. Clostridial myonecrosis, a term given to indicate true gas gangrene, it is one of the most devastating type of clostridial infections which mandates immediate and aggressive surgical intervention. It mainly involves the extremities, thighs, and shoulders.

The typical clinical course of this disease usually starts in an insidious manner that progresses rapidly, the main manifestation is severe pain not consistent with physical examination, not improving with analgesia. In addition to fever and local edema. When the infection advances, myonecrosis will be associated with necrotizing fasciitis in addition to, skin and muscle necrosis. The skin encircling the infection site will become tense and changes in color from pale to dark red with the presence of hemorrhagic bullae [12]. The condition is typically rapid in onset and progressive leading to systemic toxicity and septic shock. The diagnoses of gas gangrene remain a challenge but are based on clinical findings mainly and evidence in imaging of gas in affected areas could facilitate the diagnostic process in addition to blood tests. And the diagnosis is confirmed once a positive blood culture reveals the pathogen [7,11].

Neck Involvement

Gas gangrene around vital anatomical areas such as the neck is a life-threatening complication resulting from odontogenic infections. Its clinical picture includes fever, swallowing difficulty, or trismus. The prognosis is reportedly poor. Dental infections are one of the leading causes of head and neck infections and are usually caused by one or a combination of odontogenic issues such as pericoronitis, dental cavities, periodontitis, or a complication of dental procedures. The migration of such infections, as well as gas gangrene, depends on the affected tooth mainly in addition to, the general wellbeing of the body. Odontogenic infections are usually localized lesions that improve in response to suitable

antimicrobial agents once administered. If medical therapy fails surgical intervention is necessary and is done by incision and drainage [14,15]. However, metastatic gas gangrene to the neck in which the underlying cause was not from odontogenic infections has never been reported in the literature and the viability of such area warrants a better understanding of the predisposing factors which what we are discussing in this case.

Management

Due to the unfortunate sequel of gas gangrene and its rapidly progressive clinical course. It is crucial to promptly identify the disease and treat patients aggressively. Once gas gangrene is suspected antimicrobial therapy should be initiated immediately in addition to possible urgent surgical intervention such as exploration and debridement of affected tissue as needed. Spontaneous *C. septicum* mortality was reported to be 70% in 78 cases, 56% in 32 cases, and 80% in 241 cases in three distinct papers [7]. Fortunately, in our case, the patient didn't show any signs of infections and the computed tomography with contrast of her neck assisted in the diagnosis and we decided to manage her conservatively after extensive thorough investigation and review of the literature [7,11].

Conclusion

We are reporting metastatic gas gangrene of the neck which is a rare fulminant clinical entity in a healthy young female in which she developed during early childhood after sustaining a trauma. Further research is warranted as the condition occurred in a healthy individual involving viable structures which could lead to lethal consequences.

References

1. Srivastava I, Aldape M, Bryant A, Stevens D (2017) Spontaneous *C. septicum* gas gangrene: A literature review. *Anaerobe* 48: 165-171.
2. Alpern R, VR Dowell (1969) Clostridium septicum infections and malignancy. *JAMA* 209(3): 385-388.
3. Smith-Slatas C, M Bourque, JC Salazar (2006) Clostridium septicum infections in children: a case report and review of the literature. *Pediatrics* 117(4): e796-e805.
4. Lee H, Cho S, Lee D, Ko Y, Hyun J, et al. (2014) A Fatal Spontaneous Gas Gangrene due to Clostridium perfringens during Neutropenia of Allogeneic Stem Cell Transplantation: Case Report and Literature Review. *Infect Chemother* 46(3): 199-203.
5. Stevens D, Musher D, Watson D, Eddy H, Hamill R, et al. (1990) Spontaneous, Nontraumatic Gangrene Due to Clostridium septicum. *Rev Infect Dis* 12(2): 286-296.
6. Alba-Loureiro T, Munhoz C, Martins J, Cerchiaro G, Scavone C, et al. (2007) Neutrophil function and metabolism in individuals with diabetes mellitus. *Braz J Med Biol Res* 40(8): 1037-1044.
7. Buboltz JB, Murphy-Lavoie HM (2022) Gas Gangrene. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing.
8. Amer N, Karayanis J (2016) Metastatic clostridial myonecrosis secondary to perforated metastatic bowel cancer. *Journal of Cancer Metastasis and Treatment* 2(10): 402-406.

9. Haruka K, Harumi G, Kazutaka F, Yukiko K, Taijiro S, et al. (2017) Rapidly Progressive Fatal Gas Gangrene due to Clostridium Septicum in a Patient with Colon Cancer Revealed by Autopsy. *International Journal of Critical Care and Emergency Medicine* 3(2).
10. Thiele E, Arison R, Boxer G (1964) Oncolysis by Clostridia. III. Effects of Clostridia and Chemotherapeutic Agents on Rodent Tumors. *Cancer Res* 24: 222-233.
11. Kousa O, Essa A, Ramadan B, Aly A, Awad D, et al. (2020) Multiorgan fatal gas gangrene in the setting of clostridium septicum bacteremia: a case report. *Journal of Emergency and Critical Care Medicine* 4: 21-21.
12. Ying Z, Zhang M, Yan S, Zhu Z. (2013) Gas Gangrene in Orthopaedic Patients. *Case Rep Orthop* 2013: 942076.
13. Altemeier W, Fullen W (1971) Prevention and Treatment of Gas Gangrene. *JAMA* 217(6): 806-813.
14. Takada T, Kuriyama A (2013) Sublingual gas gangrene after dental therapy. *QJM* 107(6): 485.
15. Gamoh S, Tsuji K, Maruyama H, Hamada H, Akiyama H, et al. (2017) Gas gangrene in the deep spaces of the head and neck visualized on computed tomography images. *Oral Radiology* 34(1): 83-87.



This work is licensed under Creative Commons Attribution 4.0 License
DOI: [10.19080/GJO.2022.25.556167](https://doi.org/10.19080/GJO.2022.25.556167)

Your next submission with Juniper Publishers will reach you the below assets

- Quality Editorial service
- Swift Peer Review
- Reprints availability
- E-prints Service
- Manuscript Podcast for convenient understanding
- Global attainment for your research
- Manuscript accessibility in different formats
(Pdf, E-pub, Full Text, Audio)
- Unceasing customer service

Track the below URL for one-step submission
<https://juniperpublishers.com/online-submission.php>