

## LUDWIG'S ANGINA: AN ANALYSIS OF CASES SEEN AT THE UNIVERSITY COLLEGE HOSPITAL, IBADAN

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### ABSTRACT

**Background:** Ludwig's angina (LA) is defined as a rapidly spreading cellulitis involving submandibular, sublingual and submental tissue spaces bilaterally and simultaneously.

**Aim:** The aim of this study was to determine the causes, complications, duration of hospital stay and outcome of cases that presented within a 2-year period in the University College Hospital Ibadan and reviewing the management protocols used for such cases.

**Materials and Methods:** All cases of LA seen in the department of Oral and Maxillofacial Surgery from January 2015 to December 2016 were studied. Information retrieved from patients' case files included the aetiology, signs and symptoms at presentation, possible predisposing factors, results of laboratory investigations, duration of hospital stay and treatment outcome. Data obtained was analysed using SPSS 22.0 statistical software package (SPSS Inc., Chicago, IL, USA) to present descriptive statistics.

**Results:** There were 13 cases of LA, consisting of 7 males and 6 females with a mean age of  $47.7 \pm 16.8$  years (age range 24-80 years). An odontogenic focus was noted in all cases. Almost all patients underwent surgical treatment, which consisted of an extraction of the offending teeth as well as incision and drainage of abscesses. Length of hospital stay ranged from 1 to 30 days (Mean 10 days) with all but 2 patients surviving.

**Conclusion:** This Study recorded an 84.6% survival rate because our management is handled as a surgical emergency with early recognition and attention paid to the airway in collaboration with the Otorhinolaryngology Unit in our center.

**Keywords:** Ludwig's Angina, Underlying medical conditions, Treatment outcome

### INTRODUCTION

Ludwig's Angina also known as *morbus strangularis* and *angina maligna* was first described in 1836 by the German surgeon *Wilhelm Friedrich Von Ludwig* as a fast spreading, nearly always fatal infection involving the connective tissues of the neck and the floor of the mouth<sup>1, 2</sup>. However over the decades with better understanding of the condition coupled with advances in medical and surgical care, morbidity and mortality has been reduced drastically in many centres<sup>3,4,9</sup>. In spite of improved knowledge and better management, the potential to be fatal still remains in every case of Ludwig's angina and most centres still record a number of fatalities and morbidities although higher in the face of suboptimal management.

Regarding the aetiology, a variety of sources have been implicated, however, it most commonly arises from an odontogenic foci arising typically from the roots of the second and third molar<sup>3,6</sup> teeth with previous studies reporting 78% and 90% respectively as odontogenic foci<sup>1,7</sup>. The roots of these teeth extend

below the mylohyoid line of the mandible. This allows for progression of infection from these teeth into the submandibular space which is usually the primary space in most cases<sup>5</sup> and from there to the sublingual and submental spaces. Other aetiological sources though not as common include iatrogenic such as following procedures like frenuloplasty<sup>8</sup>, from clinical conditions like mandibular fractures<sup>9</sup>, floor of the mouth infections from an oral malignancy<sup>10,11</sup> as well as a bizarre case of a migrating fish bone.<sup>12</sup>

The bacterial aetiology is mostly polymicrobial involving both aerobic and anaerobic organisms with the spread being as a result of a synergistic action between these two groups as well as their combined virulence<sup>3</sup>. Commonly isolated microorganisms include *Streptococcus viridans*, *Staphylococcus aureus*, *Enterococcus*, *Escherichia coli*, *Bacteroides*, *Actinomyces spp* and *Pseudomonas*<sup>13</sup> with *Streptococcus* being the most common organism cultured<sup>14</sup>.

Although most cases of Ludwig's Angina are seen in immunocompetent persons, there are several factors that can predispose an individual to coming down with the disease. Examples include Diabetes Mellitus, HIV and Hypertension with Diabetes being the most common as predisposing factor as seen in a study<sup>1</sup>. Other factors include immune suppression from chronic use of steroids as well as malnutrition have also been implicated<sup>15,16</sup>.

Airway management is of prime importance in the management of these patients. Options for managing the airway include blind nasotracheal intubation, retrograde intubation, fiberoptic intubation and the creation of a surgical airway.<sup>5</sup>

Airway management alongside proper antibiotic coverage, thorough surgical drainage of the involved spaces as well as adequate supportive therapy by way of fluid resuscitation, pain control and nutritional support are the pillars upon which modern management of these cases are based upon<sup>1</sup>. These have helped reduce fatalities associated with these cases from 54% in the pre antibiotic era to around 0-8% today.<sup>3,5</sup>

The aim of this publication is to determine the causes, complications, duration of hospital stay and treatment outcome of cases of Ludwig's Angina presenting within a two year period in the University College Hospital (UCH) Ibadan whilst reviewing the management protocols used for these cases.

## MATERIALS AND METHODS

An audit was conducted of all cases of Ludwig's Angina that was seen in the Department of Oral and Maxillofacial Surgery from January 2015 - December 2016. Information retrieved from the patients' case files included the demographics, aetiology, signs and symptoms at presentation and possible predisposing factors. Laboratory investigations that were done including Full Blood Count, Electrolytes and Urea, Blood Sugar profile and Microscopy, culture and sensitivity (MCS) of all aspirates obtained. In addition, predisposing factors, complications and duration of hospital stay and treatment outcome were also noted. Those patients with comorbid conditions were jointly managed with physicians of appropriate specialty. For the sake of this study, a period of admission greater than 6 days was considered prolonged. Data analysis was done using SPSS version 22.0 statistical software package (SPSS Inc., Chicago, IL, USA) to present descriptive statistics and frequency charts.

## RESULTS

A total of thirteen cases diagnosed with LA were retrieved within the study period. There were 7 males and 6 females. (M; F ratio 1.16:1) with a mean age of  $47.7 \pm 16.8$  years (range of 24- 80 years) (Table 1).

An odontogenic focus was recorded in all cases with the commonest tooth involved being the 3<sup>rd</sup> molar as highlighted in Table 1. Multiple Teeth were involved in 2 cases.

**Table 1:** Demographics and teeth involved

	<b>Gender</b>	<b>Age</b>	<b>Teeth Involved</b>
1	Female	80	Lower 1 <sup>st</sup> Molar
2	Male	24	Lower 2 <sup>nd</sup> Molar
3	Male	25	Lower 2 <sup>nd</sup> Molar
4	Female	43	Lower 2 <sup>nd</sup> Molar
5	Male	31	Lower 3 <sup>rd</sup> Molar
6	Female	33	Lower 3 <sup>rd</sup> Molar
7	Male	38	Lower 3 <sup>rd</sup> Molar
8	Female	43	Lower 3 <sup>rd</sup> Molar
9	Male	55	Lower 3 <sup>rd</sup> Molar
10	Female	62	Lower 3 <sup>rd</sup> Molar
11	Male	64	Lower 3 <sup>rd</sup> Molar
12	Male	57	Lower 2 <sup>nd</sup> Premolar, Lower 1 <sup>st</sup> Molar, Lower 3 <sup>rd</sup> Molar
13	Female	65	Lower 1 <sup>st</sup> molar, Lower 3 <sup>rd</sup> molar

All patients presented with trismus, pain, toothache, poor oral hygiene, halitosis, swelling involving the submandibular and sublingual spaces with a raised floor of mouth and displaced tongue (Table 2).

Fever was present in eleven patients with temperatures greater than 38 degrees Celsius, whilst two patients did not present with fever.

Nine patients presented with respiratory distress out of which four had emergency tracheostomy done.

were no longer active. The extraction of teeth was done for two patients on the same day they were admitted, four patients had extractions a day after presentation and five patients had extraction done 2 days after presentation.

Nine (69.2%) patients underwent their surgical treatment under local anesthesia. Three (23.1%) patients underwent surgical treatment under general anesthesia. These cases were complicated by airway obstruction requiring emergency tracheostomy.

**Table 2:** Presenting signs and symptoms of Ludwig's Angina and frequency of occurrence

Patient	Trismus 100%	Fever 84.6%	Pain 100%	Swelling 100%	Halitosis 100%	Toothache 100%	Respiratory Distress 69.2%	Intraoral Pus Discharge in relation to swelling 38.5%	Poor oral Hygiene 100%	Raised tongue 100%
1	+	+	+	+	+	+	-	+	+	+
2	+	+	+	+	+	+	+	-	+	+
3	+	+	+	+	+	+	+	-	+	+
4	+	-	+	+	+	+	+	-	+	+
5	+	+	+	+	+	+	-	+	+	+
6	+	+	+	+	+	+	-	+	+	+
7	+	+	+	+	+	+	+	-	+	+
8	+	-	+	+	+	+	+	-	+	+
9	+	+	+	+	+	+	-	-	+	+
10	+	+	+	+	+	+	+	-	+	+
11	+	+	+	+	+	+	+	-	+	+
12	+	+	+	+	+	+	+	+	+	+
13	+	+	+	+	+	+	+	+	+	+

*Presenting signs and symptoms*

Regarding underlying illness or predisposing factors, one patient was hypertensive, two patients were diabetic, one patient was both hypertensive and diabetic and one patient was anaemic with a Packed Cell Volume (PCV) of 27.8%. (Table 3).

11 patients underwent surgical treatment which consisted of an extraction of the offending teeth and incision and drainage of abscesses where necessary as well as decompression for cellulitis with insertion of a passive rubber drain. Drains were left in place till they

Intravenous antibiotics were administered alongside with analgesics, fluids and multivitamins and Dexamethasone (Hubei Tianyao, Xiangyang Hubei, China). Antibiotics administered were Ceftriaxone (Roche, Riyadh, Saudi Arabia) (2g starting dose then 1g 12 hourly) and metronidazole (Amanta, Gujarat, India) (500mg 8hourly). These were maintained throughout the duration of the treatment. The Dexamethasone was maintained for the first 48 hours and was administered as 8mg starting dose, then 4mg every 8 hours for 48 hours.

**Table 3:** Co morbid factors and systemic findings

Co-Morbid Factor	Frequency
Diabetes Mellitus	3
Hypertension	2
Anaemia	1
Systemic Findings	Frequency
Leukocytosis	13
Uremia	2
Hypokalemia	2
MCS Growth*	1

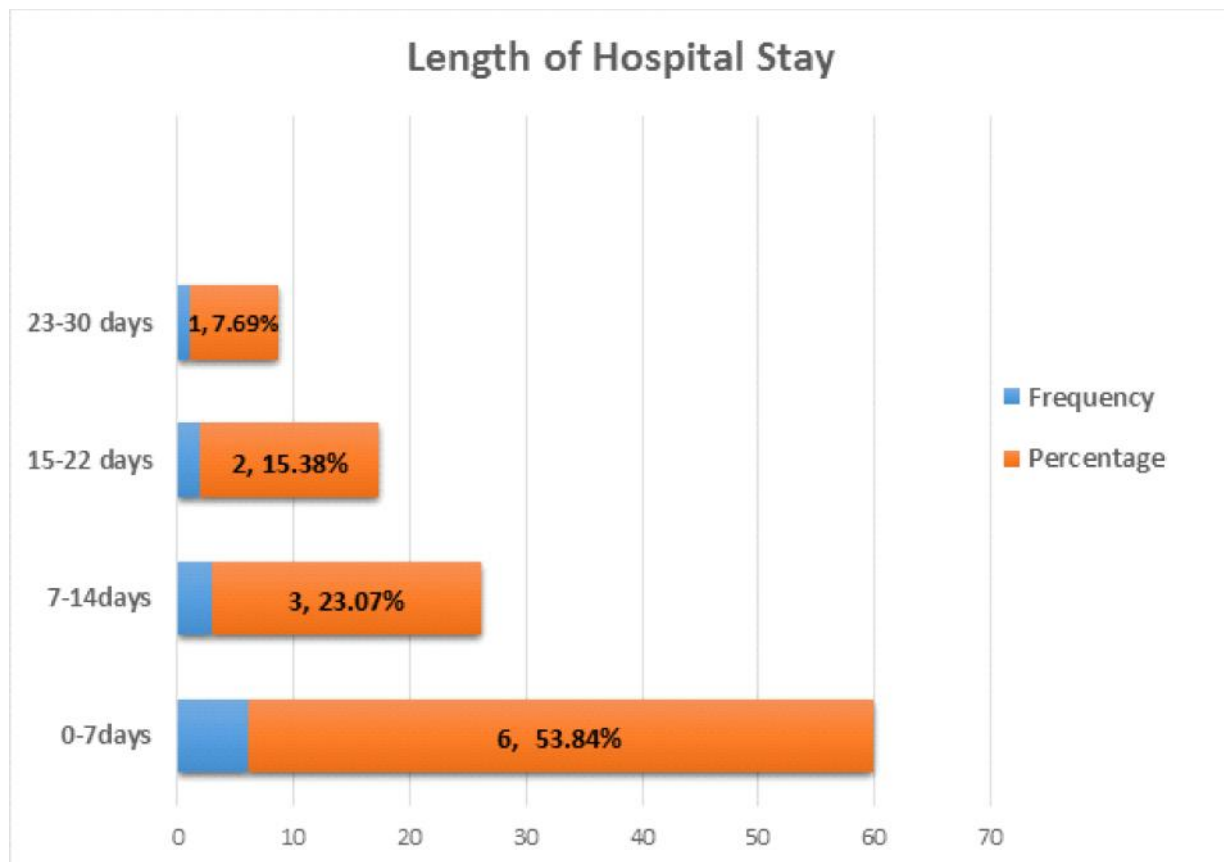
\*A positive culture was obtained in only 1 patient. This yielded an isolate of *Enterococcus faecalis*. No growth was yielded in other patients.

The different complications recorded can be found in the table below. One patient who was 8 months pregnant developed eclampsia which required an emergency caesarian delivery. This patient required mechanical ventilation in the Intensive Care Unit for 2 days.

Mean Length of hospital stay was  $10 \pm 8.2$  days (range from 1-30days). Six (46.15%) of the cases had prolonged stay in hospital longer than 6 days (Fig. 1).

Of the 6 cases who stayed longer than 6 days, three required tracheostomy because of imminent airway obstruction, two cases progressed to necrotizing fasciitis requiring further management, one developed further complications related to tracheostomy (trachea-oesophageal fistula). One patient was a known hypertensive and diabetic whose pre-existing condition was not controlled. This complicated management thereby requiring a longer stay in hospital

Regarding treatment outcome, 11 patients survived, 2 patients died. One passed on shortly after being admitted in the Accident and Emergency Unit of the hospital, the other passed on 5 days after admission.



**Fig. 1:** Length of hospital stay

**Table 4:** Surgical Drainage, Artificial Airway use, Length of Hospital Stay

Patient	Age (Years)	Extraction of offending tooth 11 (84.6%)	Incision and Drainage 9 (69.2%)	Tracheostomy (Y/N)	Length of hospital stay (days)	Systemic Disease	Disease Progression
1	31	Yes	Yes	N	3		
2	24	Yes	No	N	4		
3	38	Yes	Yes	N	6		
4	57	Yes	Yes	Y	30	Diabetes Mellitus Hypertension	Necrotizing Fasciitis
5	25	Yes	No	N	6		
6	62	Yes	Yes	N	12		
7	64	Yes	Yes	N	22	Anaemia	Necrotizing Fasciitis
8	43	Yes	Yes	Y	16		
9	33	Yes	Yes	Y	11	Hypertensive Heart Disease	Necrotizing Fasciitis
10	65	Yes	No	N	2		
11	43	Yes	No	N	12	Diabetes Mellitus Hypokalemia	Necrotizing Fasciitis Aspiration Pneumonitis Severe Sepsis
12	80	No	Yes	Y	5		
13		None		N	<1		

**Table 5:** Complications

Complication	Frequency	Percentage
Necrotizing Fasciitis	4	33.3
Severe Sepsis	2	16.7
Aspiration Pneumonitis	1	8.3
Diabetic Ketoacidosis	1	8.3
Airway Obstruction	1	8.3
Trachea-oesophageal fistula	1	8.3
Death	2	16.7

## DISCUSSION

Documented Studies have shown LA to be commoner among the male gender<sup>18,19</sup>. Botha and Colleagues reported a male to female ratio of 2.32:1 while Mahmud *et al*, reported a male to female ratio of 5.5:1. The gender gap has been postulated to be due to males being the sole breadwinner of the family in their society which often lead to neglect of dental care due to a lack of time to do so<sup>1</sup>. In our study, a narrower gap

was recorded in the male to female ratio compared to earlier studies. A possible reason for the relatively high number of females in our study may be the rising trend in female breadwinners in the Nigerian setting as highlighted by several studies<sup>20, 21</sup> and the general time constraints associated with being family breadwinners preventing them from seeking dental care in a timely manner. Ludwig's Angina tends to affect a wide age range.<sup>18,22,23</sup> The most affected age range in our study was the 4<sup>th</sup> and 7<sup>th</sup> decade of life (23.07%). This is similar with findings by Huang and colleagues in their study who reported 52.4% and 34.1% of patients being in the 5<sup>th</sup> and 7<sup>th</sup> decade respectively<sup>27</sup>. Huang *et al*. attributed it to an increase in the population of the elderly due to the progress of medical care in their environment which is a developed one. A possible reason for the distribution of patients in our study may be due to economic reasons. The high number of patients at the younger end of the spectrum (4<sup>th</sup> decade) in our study may be linked to delayed presentation due to a lack of time to present in hospital as this group is economically active. A visit to the hospital means taking time off work which this

economically active group seek to avoid. While the older age group (7<sup>th</sup> decade) may be linked to the rising trend of elder neglect by younger caregivers occasioned by economic hardship as reported by several Nigerian studies<sup>32,33</sup> leading to delayed presentation. This coupled with the relatively lower life expectancy in Nigeria<sup>38</sup> a developing country, may be responsible for the high number of patients in this older age group.

Most cases of Ludwig's angina are said to arise from an odontogenic source with some findings as high as 90% in the adult population.<sup>3,19,22,24,25</sup> This is in agreement with our study in which an odontogenic source was found in all the patients in our study. Although this is likely due to our relatively small study population, it still highlights the point that odontogenic foci are responsible for most cases of Ludwig's Angina.

It was observed in earlier studies on Ludwig's Angina, that the most common teeth involved are the mandibular 2<sup>nd</sup> and 3<sup>rd</sup> molar teeth<sup>7, 26</sup>. This is in agreement with our findings in which the mandibular 3<sup>rd</sup> molar tooth was the most involved (52.94%) followed by the 2<sup>nd</sup> molar teeth (23.53%).

Ludwig's Angina has been known to be associated with several systemic diseases such as hypertension, Diabetes, sickle cell anaemia. About a third (38.46%) of patients in our study had an associated systemic disease. This is similar to the 34.1% reported by Huang *et al.* in a review of a larger case series of 185 patients<sup>27</sup>. Diabetes was the most common co morbidity in our study, this is the same finding in other studies<sup>19,27,28,35</sup>. Diabetes is well documented in the literature as being associated with poor wound healing, compromised immunity and increased infection as seen in Ludwig's Angina<sup>36, 35, 36</sup>.

The duration of hospital stay ranged from 1 day to 30 days. Five patients (38.46%) spent less than 6 days, four of these patients underwent surgical incision and drainage and/or extraction of the offending tooth. This is less than the 93% of cases reported by Rowe *et al.* who recorded a total hospital stay of 7 days (2 days in ICU, 5 in Hospital) in their series who had equally undergone operative drainage.<sup>29</sup> In our study we observed that 6 (46.15%) patients stayed longer than 6 days. Most of these cases tended to be older subjects who developed further complications requiring tracheostomy and further management. They also had underlying systemic disease such as Diabetes Mellitus and hypertension, which tend to require prolonged management. This is in agreement with the findings of Huang *et al.* who made a similar observation in their study.<sup>27</sup>

One of the patients was 8 months pregnant at presentation who underwent a pre-term delivery via caesarian delivery. This finding is similar to that of Osunde *et al.* who observed the pre-term delivery may be likely due to overwhelming infections stemming mostly from odontogenic origin which this patient had<sup>3</sup>.

Despite the improvement in mortality rate following antibiotic therapy and surgical intervention, Ludwig's Angina is still a dangerous disease that can have a fatal outcome due mostly to the rapid airway obstruction that can occur.<sup>5,17,30,31</sup> Our study recorded two mortalities (15.38%). Both cases presented late and had uncontrolled Diabetes Mellitus as a predisposing factor with the latter case who died on the ward being complicated by necrotizing fasciitis on a background of severe sepsis, aspiration pneumonitis, acute kidney injury and diabetic ketoacidosis. These 2 cases did not undergo extraction of the offending tooth. We believe the reason for the relatively low fatalities<sup>35</sup> was because we managed the cases seen in our department as surgical emergencies with early recognition and prompt attention being paid to the airway in collaboration with the Otorhinolaryngology Unit in our centre.

## CONCLUSION

The prompt use of appropriate antibiotics, aggressive surgical drainage, adequate supportive care including appropriate fluid resuscitation, analgesia, nutritional support, airway support and management of underlying systemic conditions as indicated, possibly accounted for a more positive outcome in the management of Ludwig's Angina in our centre.

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