



OFFICIAL PUBLICATION OF
THE MINISTRY OF HEALTH,
BRUNEI DARUSSALAM

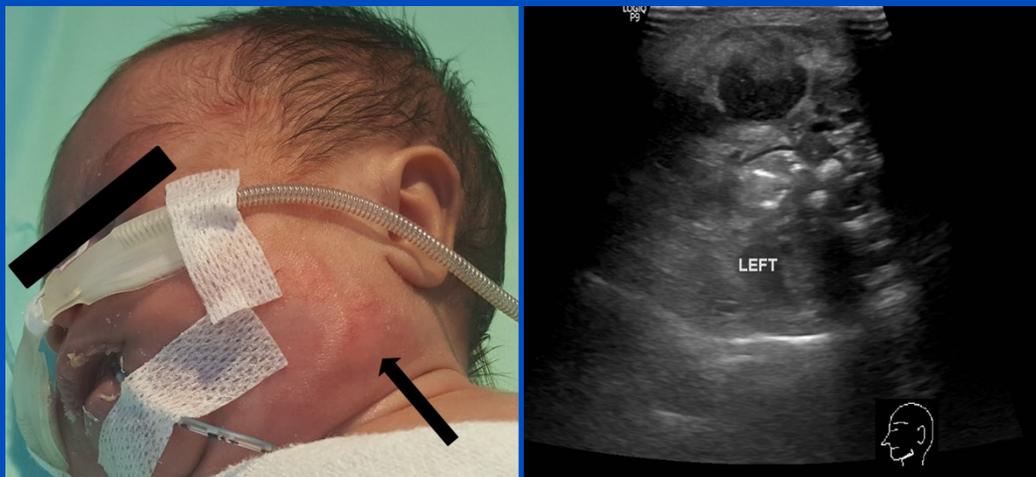
Brunei International Medical Journal

Volume 15

19 March 2019 (12 Rejab 1440H)

PRETERM NEONATAL SUPPURATIVE SUBMANDIBULAR SIALADENITIS: A CASE REPORT.

Izzah AKASHAH, Jeyasakthy SANIASIAYA, Jennifer LEE Peak Hui, Zainal Azmi ZAINAL ABIDIN
Department of Otorhinolaryngology, Hospital Selayang, Lebuhraya Selayang-Kepong, 68100 Batu
Caves, Selangor.



ABSTRACT

Suppurative submandibular sialadenitis amongst neonates is rare condition. They almost always involve the parotid gland. Herein, we described a case of a left suppurative submandibular sialadenitis in a 27-day old premature neonate which was successfully managed with incision and drainage and broad spectrum iv antibiotics. Culture and sensitivity of the pus grew staphylococcus aureus sensitive to cloxacillin. The aetiology, diagnosis treatment and possible complications of this rare condition are further discussed.

KEYWORDS: Neonate, Sialadenitis, Staphylococcus aureus, Submandibular abscess

Brunei Int Med J. 2019;15:31-34

Brunei International Medical Journal (BIMJ)

Official Publication of the Ministry of Health, Brunei Darussalam

EDITORIAL BOARD

Editor-in-Chief	William Chee Fui CHONG
Sub-Editors	Vui Heng CHONG Ketan PANDE
Editorial Board Members	Nazar LUQMAN Muhd Syafiq ABDULLAH Alice Moi Ling YONG Ahmad Yazid ABDUL WAHAB Jackson Chee Seng TAN Dipo OLABUMUYI Pemasiri Upali TELISINGHE Roselina YAAKUB Pengiran Khairol Asmee PENGIRAN SABTU Dayangku Siti Nur Ashikin PENGIRAN TENGAH

INTERNATIONAL EDITORIAL BOARD MEMBERS

Lawrence HO Khek Yu (Singapore)	Surinderpal S BIRRING (United Kingdom)
Emily Felicia Jan Ee SHEN (Singapore)	Leslie GOH (United Kingdom)
John YAP (United Kingdom)	Chuen Neng LEE (Singapore)
Christopher HAYWARD (Australia)	Jimmy SO (Singapore)
Jose F LAPENA (Philippines)	Simon Peter FROSTICK (United Kingdom)

Advisor

Wilfred PEH (Singapore)

Past Editors

Nagamuttu RAVINDRANATHAN
Kenneth Yuh Yen KOK

Proof reader

John WOLSTENHOLME (CfBT Brunei Darussalam)

Aim and Scope of Brunei International Medical Journal

The Brunei International Medical Journal (BIMJ) is a six monthly peer reviewed official publication of the Ministry of Health under the auspices of the Clinical Research Unit, Ministry of Health, Brunei Darussalam.

The BIMJ publishes articles ranging from original research papers, review articles, medical practice papers, special reports, audits, case reports, images of interest, education and technical/innovation papers, editorials, commentaries and letters to the Editor. Topics of interest include all subjects that relate to clinical practice and research in all branches of medicine, basic and clinical including topics related to allied health care fields. The BIMJ welcomes manuscripts from contributors, but usually solicits reviews articles and special reports. Proposals for review papers can be sent to the Managing Editor directly. Please refer to the contact information of the Editorial Office.

Instruction to authors

Manuscript submissions

All manuscripts should be sent to the Managing Editor, BIMJ, Ministry of Health, Brunei Darussalam; e-mail: editor-in-chief@bimjonline.com. Subsequent correspondence between the BIMJ and authors will, as far as possible via should be conducted via email quoting the reference number.

Conditions

Submission of an article for consideration for publication implies the transfer of the copyright from the authors to the BIMJ upon acceptance. The final decision of acceptance rests with the Editor-in-Chief. All accepted papers become the permanent property of the BIMJ and may not be published elsewhere without written permission from the BIMJ.

Ethics

Ethical considerations will be taken into account in the assessment of papers that have experimental investigations of human or animal subjects. Authors should state clearly in the Materials and Methods section of the manuscript that institutional review board has approved the project. Those investigators without such review boards should ensure that the principles outlined in the Declaration of Helsinki have been followed.

Manuscript categories

Original articles

These include controlled trials, interventional studies, studies of screening and diagnostic tests, outcome studies, cost-effectiveness analyses, and large-scale epidemiological studies. Manuscript should include the following; introduction, materials and methods, results and conclusion. The objective should be stated clearly in the introduction. The text should not exceed 2500 words and references not more than 30.

Review articles

These are, in general, invited papers, but unsolicited reviews, if of good quality, may be considered. Reviews are systematic critical assessments of

literature and data sources pertaining to clinical topics, emphasising factors such as cause, diagnosis, prognosis, therapy, or prevention. Reviews should be made relevant to our local setting and preferably supported by local data. The text should not exceed 3000 words and references not more than 40.

Special Reports

This section usually consist of invited reports that have significant impact on healthcare practice and usually cover disease outbreaks, management guidelines or policy statement paper.

Audits

Audits of relevant topics generally follow the same format as original article and the text should not exceed 1,500 words and references not more than 20.

Case reports

Case reports should highlight interesting rare cases or provide good learning points. The text should not exceed 1000 words; the number of tables, figures, or both should not be more than two, and references should not be more than 15.

Education section

This section includes papers (i.e. how to interpret ECG or chest radiography) with particular aim of broadening knowledge or serve as revision materials. Papers will usually be invited but well written paper on relevant topics may be accepted. The text should not exceed 1500 words and should include not more than 15 figures illustration and references should not be more than 15.

Images of interest

These are papers presenting unique clinical encounters that are illustrated by photographs, radiographs, or other figures. Image of interest should include a brief description of the case and discussion with educational aspects. Alternatively, a mini quiz can be presented and answers will be posted in a different section of the publication. A maximum of

three relevant references should be included. Only images of high quality (at least 300dpi) will be acceptable.

Technical innovations

This section include papers looking at novel or new techniques that have been developed or introduced to the local setting. The text should not exceed 1000 words and should include not more than 10 figures illustration and references should not be more than 10.

Letters to the Editor

Letters discussing a recent article published in the BIMJ are welcome and should be sent to the Editorial Office by e-mail. The text should not exceed 250 words; have no more than one figure or table, and five references.

Criteria for manuscripts

Manuscripts submitted to the BIMJ should meet the following criteria: the content is original; the writing is clear; the study methods are appropriate; the data are valid; the conclusions are reasonable and supported by the data; the information is important; and the topic has general medical interest. Manuscripts will be accepted only if both their contents and style meet the standards required by the BIMJ.

Authorship information

Designate one corresponding author and provide a complete address, telephone and fax numbers, and e-mail address. The number of authors of each paper should not be more than twelve; a greater number requires justification. Authors may add a publishable footnote explaining order of authorship.

Group authorship

If authorship is attributed to a group (either solely or in addition to one or more individual authors), all members of the group must meet the full criteria and requirements for authorship described in the following paragraphs. One or more authors may take responsibility 'for' a group, in which case the other group members are not authors, but may be listed in an acknowledgement.

Authorship requirement

When the BIMJ accepts a paper for publication, authors will be asked to sign statements on (1) financial disclosure, (2) conflict of interest and (3) copyright transfer. The correspondence author may sign on behalf of co-authors.

Authorship criteria and responsibility

All authors must meet the following criteria: to have participated sufficiently in the work to take public responsibility for the content; to have made substantial contributions to the conception and de-

sign, and the analysis and interpretation of the data (where applicable); to have made substantial contributions to the writing or revision of the manuscript; and to have reviewed the final version of the submitted manuscript and approved it for publication. Authors will be asked to certify that their contribution represents valid work and that neither the manuscript nor one with substantially similar content under their authorship has been published or is being considered for publication elsewhere, except as described in an attachment. If requested, authors shall provide the data on which the manuscript is based for examination by the editors or their assignees.

Financial disclosure or conflict of interest

Any affiliation with or involvement in any organisation or entity with a direct financial interest in the subject matter or materials discussed in the manuscript should be disclosed in an attachment. Any financial or material support should be identified in the manuscript.

Copyright transfer

In consideration of the action of the BIMJ in reviewing and editing a submission, the author/s will transfer, assign, or otherwise convey all copyright ownership to the Clinical Research Unit, RIPAS Hospital, Ministry of Health in the event that such work is published by the BIMJ.

Acknowledgements

Only persons who have made substantial contributions but who do not fulfill the authorship criteria should be acknowledged.

Accepted manuscripts

Authors will be informed of acceptances and accepted manuscripts will be sent for copyediting. During copyediting, there may be some changes made to accommodate the style of journal format. Attempts will be made to ensure that the overall meaning of the texts are not altered. Authors will be informed by email of the estimated time of publication. Authors may be requested to provide raw data, especially those presented in graph such as bar charts or figures so that presentations can be constructed following the format and style of the journal. Proofs will be sent to authors to check for any mistakes made during copyediting. Authors are usually given 72 hours to return the proof. No response will be taken as no further corrections required. Corrections should be kept to a minimum. Otherwise, it may cause delay in publication.

Offprint

Contributors will not be given any offprint of their published articles. Contributors can obtain an electronic reprint from the journal website.

DISCLAIMER

All articles published, including editorials and letters, represent the opinion of the contributors and do not reflect the official view or policy of the Clinical Research Unit, the Ministry of Health or the institutions with which the contributors are affiliated to unless this is clearly stated. The appearance of advertisement does not necessarily constitute endorsement by the Clinical Research Unit or Ministry of Health, Brunei Darussalam. Furthermore, the publisher cannot accept responsibility for the correctness or accuracy of the advertisers' text and/or claim or any opinion expressed.

PRETERM NEONATAL SUPPURATIVE SUBMANDIBULAR SIALADENITIS: A CASE REPORT.

Izzah AKASHAH, Jeyasakthy SANIASIAYA, Jennifer LEE Peak Hui, Zainal Azmi ZAINAL ABIDIN

Department of Otorhinolaryngology, Hospital Selayang, Lebuhraya Selayang-Kepong, 68100 Batu Caves, Selangor.

ABSTRACT

Suppurative submandibular sialadenitis amongst neonates is rare condition. They almost always involve the parotid gland. Herein, we described a case of a left suppurative submandibular sialadenitis in a 27-day old premature neonate which was successfully managed with incision and drainage and broad spectrum iv antibiotics. Culture and sensitivity of the pus grew staphylococcus aureus sensitive to cloxacillin. The aetiology, diagnosis treatment and possible complications of this rare condition are further discussed.

KEYWORDS: Neonate, Sialadenitis, Staphylococcus aureus, Submandibular abscess

INTRODUCTION

Infection remains most common cause of neck swelling amongst the paediatric population of which lymphadenitis prevails.¹ Apart from that, other potential aetiology of neck swelling includes congenital, inflammatory, benign and malignant lesions. Isolated submandibular sialadenitis is exceptionally rare with a reported incidence rate of 1 in 10000 admissions.^{2,3} Parotitis amongst the neonatal population has been linked with prematurity in 35–40% of cases.⁴ Similarly, in cases of isolated submandibular sialadenitis, most of the reported infants were preterm babies.² Although associated with a much lower mortality rate in the present day, in the 1970s, this condition was associated with a mortality rate of over 30% in pre-term neonates due to immature immune system.⁵ This report presents a case of an isolated suppurative sub-

immature immune system.⁵ This report presents a case of an isolated suppurative submandibular sialadenitis in a premature neonate along with discussion on the aetiology of the condition, diagnosis, treatment and follow up.

CASE REPORT

A 27-day old baby girl borned prematurely was referred for left submandibular swelling. The swelling was noticed by the attending paediatrician as the neonate has been nursed in the Neonatal Intensive Care Unit (NICU) for feeding and weight gain. Apart from the swelling, child has no fever or any other swelling elsewhere. Child was born prematurely at 30 weeks and 3 days with a birth weight of 1.19 kilogram via an uneventful spontaneous vaginal delivery. The neonate is not syndromic, with no signs of congenital malformations.

Corresponding Author: Jeyasakthy Saniasiaya, MD, MMED (ORL-HNS), Department of Otorhinolaryngology, Hospital Selayang, Lebuhraya Selayang-Kepong, 68100 Batu Caves, Selangor, Malaysia.
E-mail : shakthy_18@yahoo.com

Upon examination, child appeared active with no signs of respiratory distress. There was a palpable warm and fluctuant left submandibular swelling, measuring 5 cm x 3 cm (Figure 1). No other neck swelling or hepatosplenomegaly was noted on examination. Child was given nasal prong oxygen 1 litre per minute and was nursed using Ryles tube feeding to monitor weight gain. Blood parameters taken showed no leucocytosis or dehydration.

Ultrasound of the neck demonstrated abscess collection at the left submandibular region measuring 1.6cm x1.8cm x1.6cm, suggestive of an abscess (Figure 2). The infant was started on intravenous (iv) amoxicillin-clavulanate acid 75mg three times a day. Her parents were counselled and an incision and drainage (I&D) was performed under local anaesthesia with no sedation. Incision was made one-finger breadth from the lower border of mandible along the skin crease over the most fluctuant area and 5cc of pus was drained. The wound was dressed with povidone dressing after the procedure. Culture and sensitivity of pus grew oxacillin-sensitive *Staphylococcus aureus*. The infant subsequently recovered and completed amoxicillin-clavulanate acid iv for 7 days. Two-weeks post I&D, the wound had healed well with no



Figure 1: Left submandibular swelling measuring 5 cm x 3 cm.

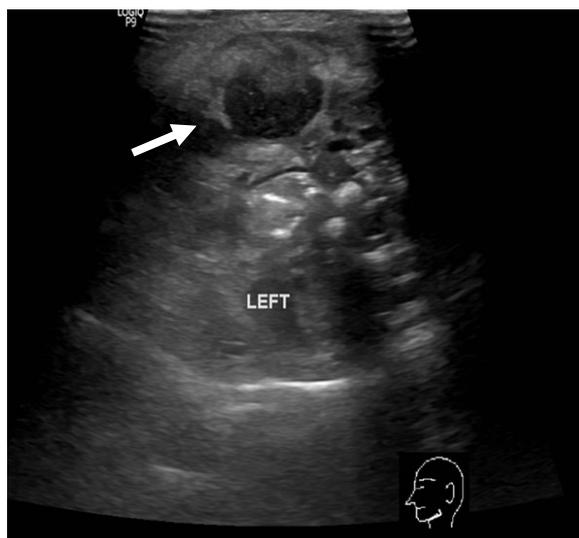


Figure 2: Ultrasound neck showing abscess collection at the left submandibular region measuring 1.6cm x1.8cm x1.6cm, suggestive of an abscess.

Staphylococcus aureus. The infant subsequently recovered and completed amoxicillin-clavulanate acid iv for 7 days. Two-weeks post I&D, the wound had healed well with no evidence of re-collection. The infant was reviewed again one month later and noted to be well with no noticeable scar and was discharged.

DISCUSSION

The most common cervical lesions within the neonatal period, are thyroglossal duct remnant and branchial cleft anomalies.¹ Infections of the salivary glands are uncommon within the neonatal period and commonly it involves the parotid gland.⁶ Submandibular suppurative sialadenitis is exceptionally rare amongst the neonates as submandibular gland produces more mucus which is bacteriostatic thus, protecting the gland from infection.^{2,7} This submandibular suppurative sialadenitis usually follows infection of the parotid gland.²

Submandibular sialadenitis was first described almost a quarter century ago.² Prior to that, submandibular sialadenitis was

to that, submandibular sialadenitis was considered secondary to parotitis. Prematurity, dehydration, prolonged gavage feeding and stasis of secretions were proposed as predisposing factors for this entity.⁸ Apart from that, presence of stones within the duct or presence of congenital malformation in the floor of the mouth could increase the likelihood of infection caused by stagnation of secretions. Only a moderate amount of dehydration is a predisposing factor for salivary sialadenitis.¹ Prolonged gavage feeding predisposes premature infants to develop submandibular sialadenitis as salivary gland stimulation is reduced which reduces ductal clearance of mucoid saliva causing obstruction and finally local inflammation.⁸

Long-term gavage feeding may predispose premature infants to submandibular sialadenitis by reducing reflex salivary gland stimulation, which can lead to reduced ductal clearance of mucoid saliva, functional obstruction and local inflammation.^{2,9} Reduced sympathetic adrenergic tone found amongst premature infants has been reported to elicit a reduction of saliva production causing obstructions of the ducts and inflammation.^{8,9} In the absence of other causes such as dehydration and congenital anomalies or tumours, it was assumed that prolonged nasogastric feeding may have contributed to the development of sialadenitis in this case. Besides prematurity and prolonged gavage feeding, the neonate in our case revealed presence of no stones, normal examination of the floor of mouth and she was also not dehydrated.

Albeit rare, cervical abscess in a newborn commonly manifests as erythematous, fluctuant swelling in the upper neck.⁹ These abscesses often arises as a sequelae of odontogenic, rhinogenic, otogenic, or aerodigestive tract infections. Common microbacteria involved with this entity includes *Staphylococcus aureus*, streptococci, *Pseudomonas aeruginosa*, *Escherichia coli* and

abscesses often arises as a sequelae of odontogenic, rhinogenic, otogenic, or aerodigestive tract infections. Common microbacteria involved with this entity includes *Staphylococcus aureus*, streptococci, *Pseudomonas aeruginosa*, *Escherichia coli* and *Moraxella catarrhalis*.^{7, 10} *Staphylococcus aureus* is the most common organism to cause neonatal sialadenitis. Amongst organisms found in isolated submandibular sialadenitis, 83% of cases were caused by *Staphylococcus aureus*.² Potential transmission of this organism includes transmission through formula or breast milk or prolonged hospitalisation.² However, our infant was only fed via Ryles tube whereby *Staphylococcus aureus* could have colonised and later possibly infected the stagnated saliva in the submandibular gland from prolonged gavage feeding.

Ultrasound remains the preferred imaging modality as it is safe, fast, cost effective, and feasible with no radiation exposure. Ultrasound helps in determining the size, vascularity, location and consistency of the mass.¹ Computed tomography scan with contrast is also useful in determining the exact location of the abscess which aids surgical drainage but exposes the neonate to significant doses of radiation.² Clinical and radiographic evidence alone is usually sufficient in diagnosing a paediatric neck mass. In our case, early diagnosis, drainage and iv antibiotics were used concurrently for successful management to reduce the progression to life-threatening complications in neonates whose immature immune systems predisposes them to serious bacterial infections.

Suppurative submandibular sialadenitis albeit rare is an important differential diagnosis in paediatrics/neonatal neck mass. The most

CONCLUSION

Suppurative submandibular sialadenitis albeit rare is an important differential diagnosis in paediatrics/neonatal neck mass. The most common predisposing factors in developing sialadenitis amongst neonates include prematurity, dehydration, prolonged gavage feeding and stasis of secretions. The successful management of this condition is reliant on early recognition and diagnosis, treatment is incision and drainage with appropriate broad-spectrum antibiotics according to culture and sensitivity, always remembering that in a neonatal population with immature immune system, such trivial infections can rapidly progress to serious infections with significant mortality.

DISCLOSURE STATEMENT

The authors reported no conflict of interest or financial liability. Consent has been obtained from both parents of the child to publish the pictures and details in this report.

REFERENCES

- 1: Skoog H, Clark DW. Submandibular neck mass in a newborn. *Proc(Bayl Univ Med Cent)*. 2017; 30:461–2.
 - 2: Subhani M, Monte DC, Roche P, Parton L. Isolated bilateral submandibular abscess in a preterm infant: a case report. *Int J Pediatr Otorhinolaryngol*. 1999;51:47–50.
 - 3: Ryan RF, Padmakumar B. Neonatal suppurative sialadenitis: An important clinical diagnosis. *Case Reports*. 2015;2015:bcr2014208535. [Accessed on 2019 February 15]. Pdf available at <https://europepmc.org/backend/ptpmcrender.fcgi?accid=PMC4442179&blobtype=pdf>
 - 4: Kaban LB, Mulliken JB, Murray JE. Sialadenitis in childhood. *Am J Surg*1978;135:570–6.
 - 5: Leake D, Leake R. Neonatal suppurative parotitis. *Pediatrics*. 1970;46:203–7.
 - 6: Tapisız A, Belet N, Çiftçi E, Fitöz S, İnce E, Doğru U. Neonatal suppurative submandibular sialadenitis. *Turk J Pediatr*. 2009; 51:180–2. [Accessed on 2019 February 15]. Pdf available at http://www.turkishjournalpediatrics.org/uploads/pdf_TJP_629.pdf
 - 6: Tapisız A, Belet N, Çiftçi E, Fitöz S, İnce E, Doğru U. Neonatal suppurative submandibular sialadenitis. *Turk J Pediatr*. 2009; 51:180–2. [Accessed on 2019 February 15]. Pdf available at http://www.turkishjournalpediatrics.org/uploads/pdf_TJP_629.pdf
 - 7: Saini N, Baghel A. Neonatal suppurative submandibular sialadenitis. *IOSR J Dent Med Sci-en*. 2013;6: 43–4. [Accessed on 2019 February 15]. Pdf available at <http://www.iosrjournals.org/iosr-jdms/papers/Vol6-issue1/I0614344.pdf>
 - 8: Lindgren C, Balihodzic-Lucovic V. Aseptic sialadenitis in preterm infants associated with long term oro-gastric tube feeding. *Eur J Pediatr* 1998; 157: 1014–6.
 - 9: Gaur A, Ambey R, Sharma A, Gupta S. Abscess mimicking pre-cervical and submandibular cystic hygroma in a newborn. *AMJ*. 2013;6:318–20. [Accessed on 2019 February 15]. Pdf available at https://pdfs.semanticscholar.org/8e63/fec34c3332e6663766e51fb9711c6e1e77c5.pdf?_ga=2.42464418.643215905.1552549189-1117972270.1552549189
 - 10: Cmejrek RC, Cotichia JM, Arnold JE. Presentation, Diagnosis, and Management of Deep-Neck Abscesses in Infants. *Arch Otolaryngol Head Neck Surg*. 2002;128:1361–4. [Accessed on 2019 February 15]. Full text article available at <https://jamanetwork.com/journals/jamaotolaryngology/fullarticle/483170>
-