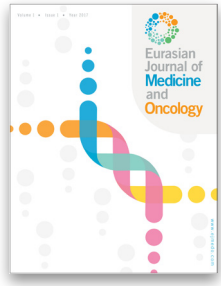
EJMO

DOI: 10.14744/ejmo.2017.42104
EJMO 2018;2(2):105-109



Case Report

Transcervical Removal of a Rare Giant Proximal Wharton's Duct Sialolith and Submandibular Gland

Muhammad Izzuddin Hamzan,[1,2] Normala Basiron[2]

[1]Reconstructive Science Unit, School of Medical Sciences, University of Science Malaysia, Kelantan, Malaysia
[2]Department of Plastic & Reconstructive Surgery, Kuala Lumpur Hospital, Kuala Lumpur, Malaysia

Abstract

Sialolithiasis, the most common salivary gland disease, is a condition in which a calculus forms in the gland, most often the Wharton's duct. Rarely, these calculi can reach several centimeters in size, and have been described as giant sialoliths in the literature. Patients diagnosed with small sialoliths can undergo conventional treatment, but those with larger sialoliths require a sialolithotomy or resection of the entire gland. The management of a salivary gland sialolith depends on its location, size, and the surgeon's experience. Presently described is a case of a giant sialolith located at the proximal part of the Wharton's duct near the hilum of the left submandibular gland, its surgical management, and a review of the current literature. To the best of our knowledge, no similar case has been reported before in the literature and this is the first case of a giant calculus in the proximal submandibular duct to be reported in the author's country of origin.

Keywords: Giant sialolith, submandibular gland, transcervical submandibulectomy, Wharton's duct

Cite This Article: *Hamzan M, Basiron N. Transcervical Removal of a Rare Giant Proximal Wharton's Duct Sialolith and Submandibular Gland. EJMO. 2018; 2(2): 105-109*

Sialolithiasis is the most frequently encountered disease of the salivary glands. It affects 1.2% of the adult population and is commonly observed among men. More than 80% of salivary gland stones develop in the submandibular gland, but they may also be located in the parenchyma and more frequently in the submandibular duct (Wharton's duct) and 50% of these are placed in the hilum or at the deep proximal part of Wharton's duct.[1, 2] Frequently, sialoliths measure <10 mm and rarely measure >15 mm in size.[1] Giant salivary gland sialoliths are stones measuring ≥15 mm and are seldom mentioned in the medical literature.[3] Giant sialoliths measuring >30 mm are extremely unusual and with only a small number of reported cases and literature review revealed only 16 published cases.[1]

Among all of the case reports and literature reviews that describe giant sialoliths in Wharton's duct, none of them elaborate on its location along the duct. Most of the giant sialoliths in Wharton's duct reported in the literature were located at the distal part of the duct or near the orifice. In this case, we reported an unusually large sialolith situated at the deep proximal part of Wharton's duct extending to the hilum of the gland at the intraglandular ductal.

Case Report

A 35-year-old gentleman presented to the Plastic & Reconstructive Surgery Unit, Hospital Kuala Lumpur (HKL) with the chief complaint of a firm mass, which was palpable in the left mandibular region for the past 5 months. He could feel the mass intra orally on the left side of the floor of the

Address for correspondence: Muhammad Izzuddin Hamzan, MD. Reconstructive Science Unit, School of Medical Sciences, University of Science Malaysia, Kelantan, Malaysia
Phone: +60 3 2615 5555 **E-mail:** drmizzud@gmail.com
Submitted Date: October 22, 2017 **Accepted Date:** December 25, 2017 **Available Online Date:** February 05, 2018
©Copyright 2018 by Eurasian Journal of Medicine and Oncology - Available online at www.ejmo.org

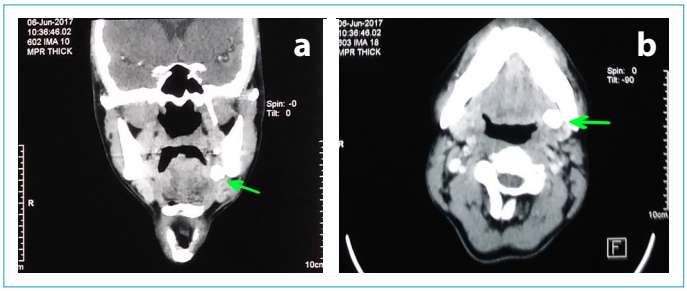


Figure 1 (a, b). Axial view **(a)** and Coronal view **(b)** of CT scan shows a calculus (Green Arrow) at the left submandibular gland.

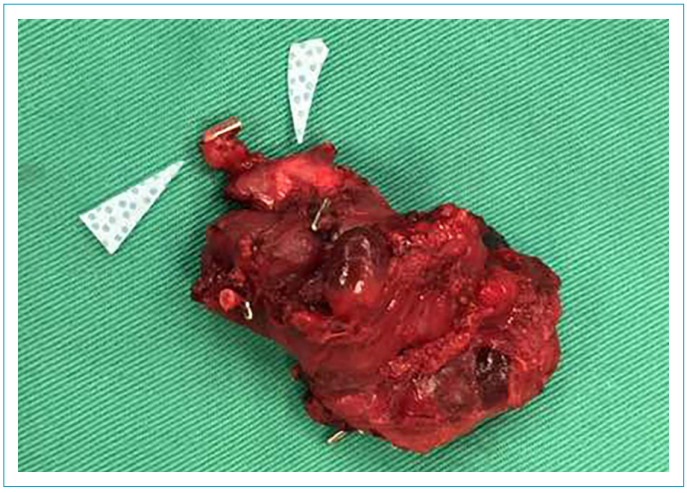


Figure 2. Left submandibular gland with its duct-cut end (Left Arrow) and calculus within the proximal duct (Upper Arrow).

mouth, which caused discomfort especially while swallowing. He had no pain, fever, chills, or other neck swelling.

Clinically, there was no obvious noticeable lump or overlying skin inflammation from the anterior and lateral view of the neck. However, there was a palpable lump measuring about 20 mm×20 mm at the left submandibular triangle which was firm, hard, and non-tender. Intra-oral examination by palpation revealed no palpable mass at the left anterior floor of the mouth. Other physical examinations were unremarkable. CT of the neck revealed a large calculus measuring 12 mm×9 mm observed at the proximal part of the left Wharton's duct with extension into the intra-glandular duct of the left submandibular gland, into the hilum (Fig. 1). The proximal left intra-glandular ducts were dilated and the left submandibular gland was slightly enlarged. The final diagnosis was sialolithiasis of the proximal left Wharton's duct with extension into the intra-glandular duct complicated with duct obstruction.

An open left submandibular gland resection was performed via transcervical approach under general anesthesia to remove both the gland and the sialolith. A direct incision was made along the inferior left mandibular line

Figure 3. The excised calculus measuring at 15 mm×15 mm.

at the left submandibular triangle while carefully avoiding the adjacent lingual nerve. No complication was encountered intra-operatively and post-operatively. The excised submandibular gland appeared to be slightly enlarged and Wharton's duct was patent distally with an obvious hard swelling at the hilar region of its proximal origin (Fig. 2). A calculus, measuring 15 mm at its widest, with an irregular round shape and hard surface was later excised (Fig. 3). He was observed at the outpatient clinic and has recovered well after the surgery with a well-healed neck scar.

Discussion

Sialolithiasis is an unusual disease and accounts for >50% of major salivary gland diseases. Males are more commonly affected than females, and children are rarely involved. Submandibular salivary glands are more commonly affected than the parotids. The sublingual or minor salivary glands are involved in only 1%–2% of the cases. Most of the Sialoliths (88%) commonly measure <10 mm in size, while only 7.6% exceed 15 mm, and the mean size is 6–9 mm.[4, 10] Sialoliths >10 mm can be reported as calculi of unusual size, and to date, the biggest salivary calculus ever reported was 72 mm in the submandibular gland of a 60-year-old man.[4]

Most submandibular sialoliths tend to manifest as single sialoliths and are located within the Wharton's duct;[5] however, unusual giant sialoliths were rarely observed within the duct as more cases were observed in the parenchyma of the gland.[6] Only a few cases of giant sialoliths in Wharton's duct have been reported around the world, and in fact none of them were located in the proximal part of the submandibular duct/hilum. In the literature review by Saluja, et al. (2012) and Babu, et al. (2001), the exact position of the giant sialoliths has not been described in detail, and most of the calculi are located at the distal part of the duct.[7, 8] Evidently, a study conducted in 2015 with 2959 calculi identified, 53% of the submandibular duct calculi were in the proximal duct with an average size of 8.5 mm and most were in Lutsman[10] group II.[9] This shows that most calculi that were found in the proximal part of Wharton's duct or hilum were not at unusually large. The shape of the sialo-

liths located in the duct is typically elongated as to follow the anatomy of Wharton's duct, while those situated within the gland or at the hilum tend to be spherical or oval as it was in this case.[10] In clinical practice, the diameter of Wharton's duct is 3–4 mm, which exceeds that of the parotid duct which is 2–3 mm, which facilitates sialendoscopic therapy with greater ease in the former than in the latter.[11] Wharton's duct can be divided into thirds of uneven length: an anterior third forward of its intersection with the lingual nerve; a posterior third, most proximal and strictly intra-glandular; and a middle third, between the other two. The anterior third part is horizontal, the two other thirds are shorter and follow a vertical course.[12]

The potential for calculus development and growth depends largely on the ability of the affected salivary duct to dilate. When a calculus is located within a duct that can dilate to permit near normal flow of saliva around the calculus, it may grow in size to become a giant calculus without producing many symptoms for a long period of time.[4] This allows the calculi in Wharton's duct to increase in size and go unnoticed as compared to Stenson's duct of the parotid gland; thus, sialolithiasis may remain asymptomatic until there is a significant obstruction of salivary flow. Some will experience xerostomia and infrequently the sensation of a gritty, sand-like foreign body in their mouth.[13]

The exact etiology of sialolithiasis is unclear. Salivary stasis, ductal inflammation, and injury promote aggregation of mineralized debris to form nidus, which ultimately initiates the formation of sialolith.[13] A number of factors are believed to be responsible in the development and growth of salivary stones in submandibular gland tissues: Wharton's duct is wider and longer than Stenson's duct, the direction of salivary flow in the submandibular duct system is in opposition to that of the force of gravity, submandibular saliva is more alkaline compared with that of the parotids, the submandibular gland secretion comprises a higher amount of mucin proteins, whereas the parotid gland secretion is totally serous, and calcium and phosphate content in submandibular saliva are greater than that in other glands.[14]

Based on 120 submandibular gland sialendoscopy studies, Marchal, et al (2001), observed the presence of a sphincter system in the first 3 cm of Wharton's duct in 90% of the cases and proposed that variation of such sphincter-like mechanism within the salivary ducts could be responsible for easier retrograde movement of oral materials.[15] Suffice to say, the predisposition to calculi and ability to tolerate expansion were what lead to a higher incidence of giant calculi in the submandibular gland.[4]

Normal saliva comprises plentiful hydroxyapatite.[13] Sialoliths consist primarily of calcium phosphate, carbonate hydroxyapatite in combination with an organic matrix of glycoproteins, and mucopolysaccharides. Small amounts of other salts such as magnesium, potassium, and ammonium are also involved in calculus formation.[16]

Careful history and examination are essential in the diagnosis of sialoliths.[17] Most of the time, if the duct near the sialolith is expandable, permitting normal secretion of saliva around the calculi, the sialolith may remain asymptomatic, therefore allowing the growth of a huge calculus over the time.[18] Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, frequently reveals a palpable calculus in most cases of submandibular calculi formation. A uniformly firm and hard gland proposes a hypofunctional or nonfunctional gland as established in the present case.[17] Imaging of the salivary gland for sialolithiasis may be accomplished with plain radiography, sialography, ultrasonography, computed tomography, and magnetic resonance imaging. Intraoral or occlusal view plain radiographs might be used to detect radio-opaque calculi and 80%–95% of submandibular sialoliths are radiopaque and can be visualized on plain radiographs.[14, 19]

The methods available for the management of submandibular sialoliths range from conservative treatment to surgical procedures. Non-invasive selections include hydration of the patient, prescription of sialogogues medication, and gland massage, all of which can be helpful in promoting salivation and result in flushing-out of the calculus. For calculi located exclusively in the duct and near the papillae, intraoral approach by using minimally invasive treatment through the manipulation of lacrimal probes and dilators to open the duct orifice in order to milk out the calculus are adequate.[17, 20, 21] Marsupialization via intraoral considered to be the best surgical option for calculi located close to the orifice of the duct.[13] The invasive, surgical approach involves an incision along the longitudinal axis of Wharton's duct directly onto the stone.[17, 21]

In regards to giant sialoliths, transoral-sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.[4] The treatment objective for giant sialoliths, as for the standard size stones, is restoration of normal salivary secretion. There are typically three ways in which patients with salivary calculi can be managed: removal through the oral cavity (intra-oral), Interventional sialoendoscopy, and resection of the gland itself. The choice rests upon the location, size, shape, number, and quality of the calculi. If possible, the giant sialolith should be removed through a minimally invasive method, via transoral sialolithotomy, to avoid morbidity linked to sialadenectomy.[18] Sialoliths that are up to 4–5 mm in diameter can be effectively removed by sialendoscopy, mainly mobile calculi that

lie freely in the lumen of Wharton's duct, and these calculi can be extracted under endoscopic control in more than 80% of the cases.[22] Statistically, almost half of the submandibular sialoliths situated in the distal third of the duct and are amenable to simple surgical release through an incision in the anterior floor of the mouth directly onto the calculi.[5, 17, 21, 23] However, sialolithotomy within the proximal duct in the so-called comma area is difficult, demanding, and may be detrimental to the lingual nerve and can be more problematic if it is a giant sialolith. Nonetheless, extended incision of the duct has been constantly preferred as a gland preserving management.[8] A prospective randomized study by Eun, et al. (2010) to compare the intra-oral and trans-cervical removal of proximal located submandibular sialolith found that the intra-oral technique have a slight advantage over the later, though the sialoliths in this study were smaller in size with mean size of 5.2 mm for the intra-oral approach and 9 mm for the trans-cervical approach.[24]

Worldwide, significant numbers of patients with giant sialoliths are treated by resection of the submandibular gland.[2] Conventionally, if the gland has been damaged by recurrent infection, obstruction and fibrosis or calculi have developed within the gland, and the structural damage acquired predisposes the gland to chronic disease and frequent impairment of the gland may necessitate removal.[17, 25] Majority of symptomatic intra-glandular sialoliths or larger sialoliths embedded in the gland which is inaccessible via a tran-soral approach entail surgical excision of the total gland.[10, 13, 17, 20] Generally, most of the sialodenectomies are performed transcervically, but it has some disadvantages; hence, minimally invasive techniques are becoming increasingly significant especially for smaller stones.[26]

In particular, intra-oral calculi removal has advantages, including patient tolerability and acceptance, a less surgery time, and the ability to be performed under both local and general anesthesia.[27] It also decreased hospital stay and proved to be less painful.[24] However, intra-oral calculi removal also has several drawbacks, including the risk of lingual nerve injury; a limited surgical field; and technical difficulty in addressing the hilar area, especially in the removal of larger and proximal submandibular calculi;[26] and the possibility of sialolithiasis recurrence.[24] Therefore, the ability to palpate the calculus, irrespective of its position or dimension, is considered to be the most important aspect in the successful trans-oral removal of the stone.[23, 27]

Foletti, et al. (2017) developed an algorithm for choosing the best minimally invasive technique for managing submandibular and parotid sialoliths, according to the size of the calculi, and their position in the excretory duct for which may be applied in smaller size calculi, not more than 10 mm of diameter.[12] Other alternative treatment for sialolithiasis includes extracorporeal shock wave lithotripsy (ESWL) and endoscopic intracorporeal shock wave lithotripsy (ISWL).[28] Larger sialoliths may be fragmented in the lumen of the duct, either mechanically, ESWL, or with a laser beam, though such a technique is impossible for calculi >6–7 mm in the submandibular hilum.[22]

There were several reports on the removal of giant sialoliths intra-orally, but all of them had calculi described within the distal portion of the Wharton's duct. Among the case reports were from Soares, et al. (2009) with 25 mm sialolith, Iqbal, et al. (2012) with 35 mm sialolith, Goyal, et al. (2013) with 50 mm sialolith, Omezli, et al. (2015) with 37 mm sialolith, Mustapha (2015) with 25 mm sialolith, Shahoon, et al. (2015) with 2 cases of giant sialolith and Gadve, et al. (2016) with 25 mm sialolith.[3, 20, 29, 30-33] However, there was one case reported of an intraoral removal of giant sialolith (25 mm) located at the proximal part of Wharton's duct but it was not mentioned whether it was extending into the intra-glandular ductal at the hilum and causing obstruction as in our case.[23]

A study by Park, et al. (2012) to identify a surgical landmark for a suitable approach to remove proximal Wharton's duct and hilum sialoliths, found that all of 74 patients had successfully underwent intraoral removal of the sialoliths with a mean size of 8 mm, which was below the size of giant sialolith.[26] In our case, an unusual large stone at proximal part of Wharton's duct which extended into intra-glandular duct at the hilum was not removable intra-orally because of a high degree of difficulty, the calculus was not palpable intra-orally, the risk of injury to adjacent important structure, and the fact that the gland which has already been damaged, was obstructed and non-functional. Therefore, based on these factors and also taking into account the surgeon's experience, a trans-cervical removal of the gland was chosen and successfully executed.

Conclusion

The management of giant sialoliths in Wharton's duct depends on its size and location and should be addressed using a technique as minimally invasive as possible, preferably via the intra-oral approach. Even in asymptomatic cases of giant sialoliths, patients should be advised for surgical involvement to avoid further or early complications. Giant sialoliths at the proximal part of Wharton's duct, deep into the hilar, are very challenging and technically demanding, which entails a more radical method by performing sialadenectomy via intra-oral or trans-cervical technique depends on case by case basis and surgeon's preference. More of such similar case studies are needed to have a broader understanding and promising data in managing of such circumstances.

Disclosures

Acknowledgements: The authors have no funding, financial relationships, or conflicts of interest to disclose.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

Authorship contributions: Concept – M.I.H.; Design – M.I.H.; Supervision – N.B.; Materials – N.B.; Data collection &/or processing – M.I.H.; Analysis and/or interpretation – N.B.; Literature search – M.I.H.; Writing – M.I.H.; Critical review – N.B.

References

1. Ledesma-Montes C, Garcés-Ortíz M, Salcido-García JF, Hernández-Flores F, Hernández-Guerrero JC. Giant sialolith: case report and review of the literature. J Oral Maxillofac Surg 2007;65:128–30. [CrossRef]
2. Schapher M, Mantsopoulos K, Messbacher ME, Iro H, Koch M. Transoral submandibulotomy for deep hilar submandibular gland sialolithiasis. Laryngoscope 2017;127:2038–44. [CrossRef]
3. Soares EC, Costa FW, Pessoa RM, Bezerra TP. Giant salivary calculus of the submandibular gland. Otolaryngol Head Neck Surg 2009;140:128–9. [CrossRef]
4. Rai M, Burman R. Giant submandibular sialolith of remarkable size in the comma area of Wharton's duct: a case report. J Oral Maxillofac Surg 2009;67:1329–32. [CrossRef]
5. Baurmash HD. Submandibular salivary stones: current management modalities. J Oral Maxillofac Surg 2004;62:369–78.
6. Kasat VO, Farooqui AA, Ladda R. Giant sialolith in the Wharton's duct. SRM J Res Dent Sci 2015;6:69–72. [CrossRef]
7. Saluja H, Kasat VO, Mahindra U. Giant Sialolith in the Wharton's duct causing sialo-oral fistula: A case report and review of literature. J Orofac Sci 2012;4:137–42. [CrossRef]
8. Lokesh BKT, Jain MK. Giant submandibular sialolith. A case report and review of literature. Int J Head Neck Surg 2001;2:154–7.
9. Sigismund PE, Zenk J, Koch M, Schapher M, Rudes M, Iro H. Nearly 3,000 salivary stones: some clinical and epidemiologic aspects. Laryngoscope 2015;125:1879–82. [CrossRef]
10. Lustmann J, Regev E, Melamed Y. Sialolithiasis. A survey on 245 patients and a review of the literature. Int J Oral Maxillofac Surg 1990;19:135–8. [CrossRef]
11. Marchal, F. Sialoendoscopy. In: Myers, EN, Ferris, RL, editors. Salivary Gland Disorders. Berlin: Springer-Verlag; 2007. p. 128–45.
12. Foletti JM, Graillon N, Avignon S, Guyot L, Chossegros C. Salivary Calculi Removal by Minimally Invasive Techniques: A Decision Tree Based on the Diameter of the Calculi and Their Position in the Excretory Duct. J Oral Maxillofac Surg 2018;76:112–8. [CrossRef]
13. Lim EH, Nadarajah S, Mohamad I. Giant Submandibular Calculus Eroding Oral Cavity Mucosa. Oman Med J 2017;32:432–5.
14. Raksin SZ, Gould SM, Williams AC. Submandibular duct sialolith of unusual size and shape. J Oral Surg 1975;33:142–5.
15. Marchal F, Kurt AM, Dulguerov P, Lehmann W. Retrograde theory in sialolithiasis formation. Arch Otolaryngol Head Neck Surg 2001;127:66–8. [CrossRef]
16. Hiraide F, Nomura Y. The fine surface structure and composition of salivary calculi. Laryngoscope 1980;90:152–8. [CrossRef]
17. Siddiqui SJ. Sialolithiasis: an unusually large submandibular salivary stone. Br Dent J 2002;193:89–91. [CrossRef]
18. Bodner L. Giant salivary gland calculi: diagnostic imaging and surgical management. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94:320–3. [CrossRef]
19. Zakaria MA. Giant calculi of the submandibular salivary gland. Br J Oral Surg 1981;19:230–2. [CrossRef]
20. Goyal L, Salim M. Rare giant submandibular gland calculus: a case report. J Evolution Med Dent Sci 2013;2:8823–6. [CrossRef]
21. Nahlieli O, Shacham R, Zagury A, Bar T, Yoffe B. The ductal stretching technique: an endoscopic-assisted technique for removal of submandibular stones. Laryngoscope 2007;117:1031–5. [CrossRef]
22. Kopeć T, Wierzbicka M, Kałużny J, Młodkowska A, Szyfter W. Sialendoscopy and sialendoscopically-assisted operations in the treatment of lithiasis of the submandibular and parotid glands: our experience of 239 cases. Br J Oral Maxillofac Surg 2016;54:767–71. [CrossRef]
23. de Santana Santos T, da Costa Araújo FA, Frota R, Caubi AF, de Oliveira E Silva ED. Intraoral approach for removal of large sialolith in submandibular gland. J Craniofac Surg 2012;23:1845–7.
24. Eun YG, Chung DH, Kwon KH. Advantages of intraoral removal over submandibular gland resection for proximal submandibular stones: a prospective randomized study. Laryngoscope 2010;120:2189–92. [CrossRef]
25. McGurk M, Esudier M. Removing salivary gland stones. Br J Hosp Med 1995;54:184–5.
26. Park HS, Pae SY, Kim KY, Chung SM, Kim HS. Intraoral removal of stones in the proximal submandibular duct: usefulness of a surgical landmark for the hilum. Laryngoscope 2013;123:934–7.
27. Krishnan B, Gehani RE, Shehumi MI. Submandibular giant sialoliths-2 case reports and review of the literature. Indian J Otolaryngol Head Neck Surg 2009;61:55–8. [CrossRef]
28. Iro H, Schneider HT, Födra C, Waitz G, Nitsche N, Heinritz HH, et al. Shockwave lithotripsy of salivary duct stones. Lancet 1992;339:1333–6. [CrossRef]
29. Iqbal A, Gupta AK, Natu SS, Gupta AK. Unusually large sialolith of Wharton's duct. Ann Maxillofac Surg 2012;2:70–3. [CrossRef]
30. Omezli MM, Ayranci F, Sadik E, Polat ME. Case report of giant sialolith (megalith) of the Wharton's duct. Niger J Clin Pract 2016;19:414–7. [CrossRef]
31. Mustapha C. Giant Submandibular Duct Calculus: A Case Report . Surgery Curr Res 2015;6:1.
32. Shahoon H, Farhadi S, Hamedi R. Giant sialoliths of Wharton duct: Report of two rare cases and review of literature. Dent Res J (Isfahan) 2015;12:494–7. [CrossRef]
33. Gadve V, Mohite A, Bang K, Shenoi SR. Unusual giant sialolith of Wharton's duct. Indian J Dent 2016;7:162–4. [CrossRef]