Unusual Large Submandibular Sialolithiasis (Stone) Report of One Case

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Abstract

Sialolithiasis is one of the most frequent disease regarding salivary glands and considered as an important cause of salivary gland dysfunction. The submandibular gland is most frequently affected. The aims of this article were to report the case of patient suffering of sialolithiasis caused by giant salivary gland calculi occurring in the Wharton’s duct, and represent the results of the literature on submandibular giant sialolith. This describes giant sialolith of submandibular gland in the lights of the literature. The comparison with the literature showed that sialolith of large size is extremely rare.

Keywords: Sialolith; Stone; Submandibular Gland

Introduction

Sialolithiasis (salivary calculi or salivary stones) is one of the most frequent disease regarding salivary glands and considered as an important cause of salivary gland dysfunction. The submandibular glands, has long and tortuous duct with a narrow orifice compared to the main part of the duct, is the most frequently affected because of its anatomical location [1,2]. Less commonly the parotid gland or rarely the sublingual gland or a minor salivary gland may develop salivary stones [3-6]. Intra-ductal stones are more common than intra-glandular stones [3-7]. Male predominance has been reported in the literature, however, Alcure., et al. reported that females being involved more often in their study [8]. The stones commonly affect middle-aged patients ranged within 42 to 58 years [1,8]. Alkaline saliva rich in mucin also contributes to factors assist stone formation [9].

Review of Literature

McCullom., et al. reported an 8-year-old black female with a curved, elliptical, stone about 10 mm long and 3 - 4 mm in diameter [10]. The stone located between the apical portion of unerupted tooth and the inferior border of the right mandible.

Dalkiz., et al. reported a 21-year-old man with circumscribed stone approximately 15 x 10 x 5 mm [11]. Gonçalves., et al. reported a 52-year-old Caucasian female with stone measured 22 mm in length with the colour varying from white to yellow [12]. Siddiqui reported a 52-year-old female with stone in the right submandibular duct measured 30 mm long along its greatest length [13].

Batori., et al. reported a 64-year-old man with the complaint of pain and swelling, correlated with meals, in his left submandibular region that had been present for 4 months. Head radiograms and ultrasonography revealed a sialolith of 13 mm in length and 7 mm in diameter at its widest portion [9].

Waśkowska, et al. reported a case of a 38-year-old man with right submandibular gland calculus showing non-typical (fang-like) shape and the size of a 35 mm [14].

Batzakakis, et al. reported a 70-year-old woman complained of intermittent episodes of moderate to severe pain and swelling in the submandibular region for a week. The gland was solid and strongly adherent to the neighbouring tissues and the stone was yellow with nodular surface. The ultrasound examination revealed a 14 mm size oval-shaped stone, inside the right submandibular gland’s substance with an irregular arrangement of gland parenchyma and without swelling of local lymph nodes [15].

Biddle and Arora reported a 48-year-old man with the stone measuring 26 x 21 x 15 mm. The calculus had a granular surface, and it was round with tan colour. It surrounded by white fibrous tissue. There was unremarkable lobulated yellow salivary gland tissue near the calculus [16].

Ayaz, et al. reported 53-year-old male with left submandibular ductal stone measured 34 x 16 mm [17].

Krishnan, et al. reported two cases of submandibular stone. The first case was a 41-year-old Sudanese man reported to the Department of Oral and Maxillofacial Surgery, following referral by a general dental practitioner, for management of a firm mass in the anterior part of the left side of the floor of the mouth. The stone measured to be 34 mm along its greatest length. Case 2; a 32-year-old Libyan woman reported to the Department of Oral and Maxillofacial Surgery, with a mass in the left side of the floor of the mouth. The stone measured to be 25 mm along its greatest length [18].

Alkurt and Peker reported two cases. The first case was a 45-year-old male, for a whitish mass in the anterior part of the right side of the oral floor. The lesion size was 28 × 8 × 4 mm. The second case was a 65-year-old male presented replace his removable prosthesis. The lesion removed surgically; the stone size was 31 × 10 × 7 mm [19].

Sathish, et al. reported a 13-year-old female complained of pain and swelling in the lower left jaw. On examination, there was obvious swelling about 40 x 30 mm in the left lower submandibular region extending about 1 cm from the midline to the angle of mandible posteriorly [20].

Miloglu, et al. reported four cases of submandibular stone. Case 1; a 59-year-old female was referred to the clinic for tooth loss and prosthetic requirement. During routine radiographic examination, a large radiopacity detected under the right-angle region of the mandible. The giant sialolith measured 35.2 × 14.0 × 12.3 mm. Case 2; a 59-year-old male was referred for a hard mass on the left floor of the mouth. The giant sialolith measured 22.0 × 9.5 × 11.2 mm. Case 3; a 48-year-old female referred to the clinic for dental caries and tooth loss. The bilateral sialoliths observed on the coronal, axial, and cross sections in the right and left submandibular regions, which measured 4.5 × 6.8 × 4.8 mm and 7.5 × 7.2 × 6.1 mm, respectively. Case 4; a 45-year-old female was referred to the clinic for periodontal problems. On routine panoramic radiograph, a radiopacity noticed under the left-angle region of the mandible and sialolithiasis was suspected. The sialolith measured 7.1 × 7.5 × 5.5 mm [21].

A 53-year-old man referred to the Oral and Maxillofacial Surgery Department at Damascus General Hospital. He complained of a large mass in the left side of the floor of the mouth. A CT scan showed a 3.32 × 1.14 cm stone blocking the submandibular gland duct [22].

Tanushri, et al. reported a 60-year-old male had a stone, 6 × 9 mm in size at left Wharton’s duct [23]. Omal and Mathew reported a 62-year-old woman with oval shaped 3 × 2 cm stone at left Wharton’s duct [24]. Shah, et al. reported a 61-year-old man, his salivary gland measured 4.8 cm in its largest dimension, and the enclosed calculus measured 4.0 cm [25].

Oteri, et al. report two cases; case 1 a 40-year-old female, had an oval sialolith about 2 cm long and 0.6 cm large located within the right Wharton’s duct. Case 2 a 51-year-old female, had a rounded sialolith with a diameter of 1.5 cm. located within the left submandibular duct [26].

Case History

A 55-year-old man referred to private Dental Clinic in Dongola city, Sudan. He complained of swelling and pain in the left submandibular area for 11 months. He had a history of left submandibular swelling occurring with meals. The past medical history of the patient was

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unremarkable. Bimanual examination and palpation of the swollen area corresponding to the anatomical location of the left submandibular salivary gland duct indicated that the mass was mobile and firm without tender. The oral floor was swollen. No cervical lymphadenopathy was present and neck movement was normal. Lateral skull X-ray revealed a large calcified mass in that area (Figure 1).

Figure 1: Lateral skull X-ray displays the calcified mass.

After explaining to the patient, the diagnosis and the procedure risks and benefits, informed consent obtained and signed by the patient. The stone excised via an incision in the floor of the mouth and directly over the palpable mass under local anaesthesia. The calculus was irregular in shape and had a rough, irregular surface measuring 42 x 28 x 13 mm (Figure 2) and yellowish coloured.

Figure 2: The calcified mass (stone).
Table 1: Summary of previous reported cases of submandibular stone.

<table>
<thead>
<tr>
<th>No.</th>
<th>Author(s)</th>
<th>Sex of Patient</th>
<th>Age of Patient</th>
<th>Site of stone</th>
<th>Size of stone (mm)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>McCullom., et al.</td>
<td>Female</td>
<td>8</td>
<td>Right submandibular duct</td>
<td>10 x 3.5</td>
</tr>
<tr>
<td>2</td>
<td>Dalkiz., et al.</td>
<td>Male</td>
<td>21</td>
<td>Right submandibular duct</td>
<td>15 x 10 x 5</td>
</tr>
<tr>
<td>3</td>
<td>GoncÊalves., et al.</td>
<td>Female</td>
<td>52</td>
<td>Left submandibular duct</td>
<td>22</td>
</tr>
<tr>
<td>4</td>
<td>Siddiqui</td>
<td>Female</td>
<td>52</td>
<td>Right submandibular duct</td>
<td>30</td>
</tr>
<tr>
<td>5</td>
<td>Batori., et al.</td>
<td>Male</td>
<td>64</td>
<td>Left submandibular duct</td>
<td>13 x 7</td>
</tr>
<tr>
<td>6</td>
<td>WaÊskowska., et al.</td>
<td>Male</td>
<td>38</td>
<td>Right submandibular duct</td>
<td>35</td>
</tr>
<tr>
<td>7</td>
<td>Batzakakis., et al.</td>
<td>Female</td>
<td>70</td>
<td>Right submandibular gland</td>
<td>14</td>
</tr>
<tr>
<td>8</td>
<td>Biddle and Arora</td>
<td>Male</td>
<td>48</td>
<td>Right submandibular duct</td>
<td>26 x 21 x 15</td>
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<tr>
<td>9</td>
<td>Ayaz., et al.</td>
<td>Male</td>
<td>53</td>
<td>Left submandibular duct</td>
<td>34 x 16</td>
</tr>
<tr>
<td>10</td>
<td>Krishnan., et al.</td>
<td>Male</td>
<td>41</td>
<td>Left submandibular duct</td>
<td>34</td>
</tr>
<tr>
<td>11</td>
<td>Alkurt and Peker</td>
<td>Female</td>
<td>32</td>
<td>Left submandibular duct</td>
<td>25</td>
</tr>
<tr>
<td>12</td>
<td>Male</td>
<td>45</td>
<td>Right submandibular duct</td>
<td>28 x 8 x 4</td>
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</tr>
<tr>
<td>13</td>
<td>Sathish., et al.</td>
<td>Female</td>
<td>13</td>
<td>Left submandibular duct</td>
<td>10</td>
</tr>
<tr>
<td>14</td>
<td>Male</td>
<td>65</td>
<td>Right submandibular duct</td>
<td>31 x 10 x 7</td>
<td></td>
</tr>
<tr>
<td>15</td>
<td>Miloglu., et al.</td>
<td>Female</td>
<td>59</td>
<td>Right submandibular duct</td>
<td>35.2 x 14 x 12.3</td>
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<tr>
<td>16</td>
<td>Male</td>
<td>59</td>
<td>Left submandibular duct</td>
<td>22 x 9.5 x 11.2</td>
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</tr>
<tr>
<td>17</td>
<td>Female</td>
<td>48</td>
<td>Bilateral: right duct</td>
<td>4.5 x 2.8 x 4.8</td>
<td></td>
</tr>
<tr>
<td>18</td>
<td>Left duct</td>
<td></td>
<td>7.5 x 2.7 x 6.1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>19</td>
<td>Female</td>
<td>45</td>
<td>Left submandibular duct</td>
<td>7.1 x 7.5 x 5.5</td>
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<tr>
<td>20</td>
<td>Abdeen and Al khen</td>
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<tr>
<td>21</td>
<td>Omal and Mathew</td>
<td>Female</td>
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<td>Left submandibular duct</td>
<td>30 x 20</td>
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<td>Shah., et al.</td>
<td>Male</td>
<td>61</td>
<td>Left submandibular gland</td>
<td>40</td>
</tr>
<tr>
<td>23</td>
<td>Female</td>
<td>40</td>
<td>Right submandibular duct</td>
<td>20 x 6</td>
<td></td>
</tr>
<tr>
<td>24</td>
<td>Oteri., et al.</td>
<td>Female</td>
<td>51</td>
<td>Left submandibular duct</td>
<td>15</td>
</tr>
<tr>
<td>25</td>
<td>Male</td>
<td>45</td>
<td>Left submandibular duct</td>
<td>42 x 28 x 13</td>
<td></td>
</tr>
</tbody>
</table>

Discussion

Sialolithiasis is the most common disease of salivary glands [1,2]; the submandibular gland is most frequently involved [3-6]. Intra-ductal calculi are more common [3-7,9-14,16-22,24,25] when compared to intra-glandular [15,23]. Male predominance has been found in the majority of the studies in the literature, whereas few authors found females affected more often in their studies [8,10,12,13,15,18,20,21,23,24].

The lesion most commonly affects middle-aged patients [1,8], the age range being 40 - 70 years [9,12,13,15-19,21-26]. While children rarely affected [10,11,14,18,20].

The size of the stone showed large range of variation, the length ranged between (4.5 - 40) mm [21,25] and the diameter ranged between (3.5 - 28) mm [10,16], the current case measured 42 x 28 x 13 mm, that the largest stone reported. The shape varied from curved, elliptic [10], fang-like [14], oval [15,24], round [16,26] or irregular as the current case. The colour varied from white to yellow [12,15] and tan [16].

Conclusion

Sialolithiasis is one of the most frequent diseases regarding salivary glands and considered as an important cause of salivary gland dysfunction. The submandibular glands are most commonly affected followed by parotid, sublingual and minor salivary glands. The stones commonly affect middle-aged patients. The size of the stone showed large range of variation, the length ranged between (4.5 - 40) mm and the diameter ranged between (3.5 - 28) mm according to literature.

Bibliography


