Developmental dysplasia of the hip screening during the lockdown for COVID-19: experience from Northern Italy

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Abstract
Purpose: Developmental dysplasia of the hip (DDH) ultrasound screening (USS), which is usually performed in Italy as an outpatient, was halted for an indefinite period in most centres during lockdown. The aim of this study was to analyze the effect of COVID-19 on DDH-USS, in two paediatric orthopaedic centres in one of the most critical areas of the Western World.

Methods: An academic teaching hospital and paediatric trauma centre (T) and a University hospital and DDH referral centre (H), classified as national COVID-19 hubs, were involved. Graf’s method was applied in both centres. In T, paediatricians directly referred only patients with delayed DDH-USS or suspicious unstable hips; in H, paediatricians or parents could directly refer to the screening service.

Results: The mean age of the 95 patients (190 hips) who were referred for DDH-USS in T, was 3.85 months (0.1 to 7.4); 175 were type I, nine were type IIa (+ and -), five type D and one type IV. In H, the screened patients in 2020 were only 78% of the same period in 2019. A total of 28 patients with 32 hips (8 IIb, 5 IIc, 8 D, 11 III) had late diagnosis at a mean age of 114 days (96 to 146). In the same period in 2019 only eight patients with 11 hips (8 IIb, 1 D, 1 III, 1 IV) at a mean age of 142 days (92 to 305) had late diagnosis.

Conclusion: DDH-USS was the only screening in newborns which halted during lockdown. Few centres, which still performed diagnosis and treatment, were overloaded causing a delay in DDH management.

Level of evidence: IV.

Introduction
On 10 March 2020, the Italian Government declared a national lockdown in response to the COVID-19 pandemic. This marked the beginning of the ‘phase 1’, which lasted from 10 March 2020 to 03 May 2020. During that period, SARS-CoV-2 was spreading widely and could not be contained, moreover the resources of the national health system were not enough and a triage for disasters was adopted, forcing authorities to reallocate economic and health resources. As already described in detail by the European Societies for Orthopedics and Traumatology as well as in Singapore, only patients requiring urgent or early orthopaedic care (such as trauma or musculoskeletal tumours) were admitted to the hospital and outpatient duties were restricted as far as possible. At the beginning, the lockdown’s duration was unknown, and in the end, it lasted almost two months. The spread of COVID-19 during phase 1 was uneven and spotty, most spots being in Northern Italy and particularly in Lombardy, Emilia-Romagna and Veneto. Lombardy, a region with almost ten million inhabitants became at that time the biggest COVID-19 spot in the world. Interestingly, the most affected area was not the local biggest metropolitan capital (Milano), but the densely populated countryside area.

In Italy, the screening programme for developmental dysplasia of the hip (DDH), which is usually performed as outpatient, was halted and postponed for an indefinite period in most centres. In this European region the expected incidence of DDH is high and lies between 1.6% and 8%. Patients are referred by their paediatrician to dedicated radiologist, paediatrician or paediatric orthopaedists.

The aim of this study was to analyze the experience of DDH screening during lockdown for COVID-19, in two paediatric orthopaedic centres located in Bergamo and Milano, which at that time were amongst the most critical areas in the Western world.
Materials and methods

In this retrospective multicentric study, data concerning DDH clinical and ultrasound screening (USS) during the lockdown in Northern Italy (from 10 March 2020 to 03 May 2020) and during all the year 2020 were analyzed. Patients’ epidemiological data and results of the screening were retrieved from medical charts. Two centres were involved: one is an academic teaching hospital and paediatric trauma centre (T) and the other one is a university hospital and DDH referral centre (H). Both have a paediatric orthopaedic service and were classified as national COVID-19 hubs during lockdown. To quantify the birth volume of both centres, in T there are on average 3500 births per year, in H 2100 births per year.

A different logistical approach was adopted. In centre T, the DDH-USS was conducted in-hospital, in the outpatient clinic during dedicated shifts; the paediatricians in the territory directly referred only patients with risk factors (breech presentation, positive family history, syndromes), suspicious unstable hips (Ortolani positive) and with delay in DDH-USS. In H, the paediatrician and/or parents could directly refer to the screening service; the DDH-USS was also conducted in a private outpatient clinic outside the hospital area for parents who chose this modality.

Graf’s method14 was used for the screening both in T and H. For the purpose of this study, patients were defined DDH-pos if they had at least one pathological hip (Ilb or c, D, III, IV according to Graf’s classification) or immature hips (Ilb+/-; even if not pathological, they represent potentially a low-grade dysplasia, requiring a second check), otherwise they were defined DDH-neg (Ia or b).

Patients who underwent the first orthopaedic evaluation for DDH-USS during lockdown in centre T and H were included. Patients already followed for DDH or evaluated for a second check were excluded.

This study was performed in accordance with the institutional review board of both centres (T and H), data were collected from registries and charts from both institutions in aggregated and anonymous form.

In total, during phase 1 of the COVID-19 lockdown, in T out of the 95 patients (190 hips) included, 57% were female. In H, 199 patients (398 hips) were screened and 49% were female; in the whole of 2020, 1083 patients (554 female and 529 male) underwent DDH-USS.

Statistical analysis

Sample distributions were tested for normality with the Kolmogorov-Smirnov test. Accordingly, data were described as appropriate (t-distribution or ranking data). Statistical analyses were computed with Microsoft Excel (Microsoft Corporation. Microsoft Excel, Redmond, US) and GraphPad Software (San Diego, US) Student’s t-test.

Results

In T, the mean age at screening was 3.85 months (0.1 to 7.4). In total, 10/95 patients had pathological findings or needed a second check, 4/10 bilaterally. In all, 6/10 started conservative therapy for DDH. A total of 175/190 were Graf type I (a and b), 9/190 type Ilb (+ and -) and underwent a second check, 5/190 type D and 1/190 type IV. The screening of two patients was delayed two weeks, as their parents had symptoms compatible with COVID-19. One mother suffered COVID-19 close to delivery and underwent an urgent caesarean but the newborn had no problems; no one else had COVID-19 symptoms. The relationship between age at screening and appointment date for DDH-USS during lockdown is plotted in Figure 1: out of 95, 71 (74%) of the patients were more than three months old at screening. Figure 1 also shows a dramatic reduction of the referred children during the first weeks of lockdown and a direct relationship between age at DDH-USS and lockdown duration.

In T, DDH-pos patients were 0.9 months old (0.1 to 2.7) at screening while DDH-neg patients were 4.2 months (0.1 to 7.4); the difference of the averages between DDH-pos and DDH-neg was 3.26 months (95% confidence interval 4.06 to -2.46). This result is biased by the fact that patients were selected by paediatricians based on clinical evaluation and risk factors, moreover, the appointment was given according to paediatric orthopaedic surgeon’s subjective perception of urgency during contact with paediatricians more than just the order of presentation. Most screening examinations were performed towards the end of the lockdown (Fig. 2).

In H, 1083 patients (2166 hips) were screened. The DDH-USS was conducted in an outpatient clinic also outside the hospital area. The number of patients who underwent DDH-USS and the age distributions in 2020 and 2019 are compared in Figure 3. The difference in age at presentation for DDH-USS calculated between 2019 and 2020 is statistically significant (p < 0.0001). At the end of the year 2020, US hip examinations decreased by 22% compared with 2019, with 1083 and 1401 patients examined respectively. The incoming second COVID-19 wave and the reduction of appointments given to the patients due to the new social distancing rules introduced after the first COVID-19 wave by the regional health authority may have determined this decrease. Beginning with phase 1 of the COVID-19 lockdown (10 March 2020) and until the end of the year, the following hip types of DDH with a late diagnosis were collected in 28 patients at a mean age of 114 days (96 to 146): eight of type Ilb, five type IIC, eight...
Fig. 1 Relationship of age at screening and screening date during lockdown in centre T.

Fig. 2 Relationship between age at screening and screening date during lockdown in centre T, grouped in developmental dysplasia of the hip (DDH)-neg and DDH-pos (for the purpose of this study, patients were defined DDH-pos if they had at least one pathological hip (IIb or c, D, III, IV according to Graf’s classification) or immature hips (IIa+/-; even if not pathological, they represent potentially a low-grade dysplasia, requiring a second check), otherwise they were defined DDH-neg (Ia or b)).

type D and 11 type III. In the same period in 2019 only eight patients with 11 hips (eight type IIb, one type D, one type III and one type IV) at a mean age of 142 days (92 to 305) had a late diagnosis.

In both centres the appointments were given to avoid patients’ overlapping and crowding; for this reason, the time dedicated for each DDH-USS was about 30 minutes (including the time of clinical evaluation itself and for use of personal protective equipment (PPE)). It took place in dedicated areas, avoiding inter-patient contact (Fig. 4). Proper PPE was used as recommended by the World Health Organization\textsuperscript{15} and other authors.\textsuperscript{16-18}
Fig. 3 Comparison between 2019 (blue) and 2020 (orange). The graph shows the distribution in the two different periods of number of patients who underwent developmental dysplasia of the hip ultrasound screening (DDH-USS) (y-axis, days) and age at presentation for DDH-USS (x-axis, days), with statistical significance (p < 0.001).

At the time of writing this article, detected DDH are now mature hips or are still in treatment with none needed to switch to open reduction.

Discussion

According to regional and national requirements, every elective and non-urgent activity was suspended in Lombardy during lockdown for the first COVID-19 wave in 2020. DDH-USS was the only screening for newborns which halted during lockdown. The experience of two centres for DDH diagnosis and treatment, which became during that period hubs hospitals for COVID-19, are reported.

Farrell et al from Canada, suggested suspending DDH screening during lockdown, as other non-urgent procedures. Indeed, DDH is not an urgent situation and the aim is understandably to avoid unnecessary exposure both for patients and caregivers. The risk of a late diagnosis must be balanced with the risk of contamination of patients and parents with SARS-CoV-2 during the clinical evaluation.

In our experience, Farrell et al’s suggestion cannot be applied in Italy and, more in general, in countries with a similar setting for DDH. Firstly, the duration of lockdown was unknown, secondly the delay must be calculated not for children born during lockdown, but for those already waiting for the DDH-USS before the instigation of the lockdown. Much literature has already been published on this topic. Schaeffer et al cited the good results of San- kar et al, justifying the possibility of postponing DDH screening during the pandemic, accepting a late diagnosis and a delayed treatment. The authors reported actually, in infants at a median age of eight months, a failure of closed reduction in 9% of the cases and a rate of 25% of avascular necrosis after closed reduction. Schaeffer et al also recognized the risks of the long-term impact on the patient’s quality of life after late treatment.

In 54 days of lockdown and in the following part of the year we observed in both centres together 34 patients
with late diagnosed DDH in patients older than three months. The high rate of DDH cases referring to H in this period is explained by the fact that H is a DDH referral centre. Patients went to H directly or after a previous clinical or US suspected diagnosis of DDH, performed elsewhere.

The number of performed procedures decreased also after the first lockdown; this could be due to more reasons. On one side, the population was comprehensively scared by COVID-19-hub hospitals, suddenly made world famous by transparent reports in the media;\textsuperscript{25,26} thus, families tried to avoid a newborn screening in these centres, although dedicated paths for COVID-19 patients had been created. In other cases, families did not find an appointment on time, due to the reduction of available places; more than one gave up, feeling that the screening was not so important, as it is not mandatory as other newborn screenings.\textsuperscript{19} Even if the situation improved substantially in June and July, the number of performed procedures did not recover in comparison with 2019. This could be explained by the above mentioned reasons and by the fact that the availability of appointments was limited by the assemblage and social distancing policies. Before the pandemic, 12 newborns per hour were scheduled, whilst during the lockdown only two or four could be done per hour; this reduced substantially the appointments on offer. Some parents must turn to private practice offices, which re-opened with the above-mentioned limitations (in Italy, mandatory newborn screenings are free of charge for families). In 2020 in H the screening procedures dropped by 22%, with a peak of -37% in March.
As above mentioned, only patients with risk factors or with delay in DDH-USS were referred to T during lockdown, whilst before 2020, only previously screened patients with a pathological or suspected DDH were examined. Thus, different cohorts of patients do not allow a correct comparison between 2019 and 2020, representing a limit of this study. Even for patients referred to H, as above described, the prevalence in cohorts of 2019 and 2020 are not comparable; despite Figure 3 showing how age is statistically significant.

Another limit of this study is that at the time of writing this article, patients are still in treatment and consequences of late diagnosis are not available for all patients, although the effects of a late treatment are well known independently of its cause. An incomplete acetabulum development with residual radiological signs of dysplasia represents a potential risk which can only be presumed and not quantified for patients involved in this study.

During the rearrangement for the pandemic of both the Orthopaedic and Traumatology Departments and the Territorial Health Care System, an effort should be made to keep the DDH-USS going.

The two different DDH-USS approaches in T and H during the lockdown described above proved to be manageable as an acute solution, but improvable, particularly for families who do not have access to private healthcare systems.

According to our experience, to balance the risk of SARS-CoV-2 contamination with the risk of DDH late diagnosis, two possible scenarios are suggested:

a) Performing DDH-USS in a dedicated outpatient service.

b) Performing DDH-USS as an inpatient service, as neonatal screening in neonatal unit care before discharge.

Both strategies, (a) and (b) can be combined, avoiding patients’ centralisation in hospitals, overall if those are COVID-19 hubs. Moreover, an overload of the service must be considered, due to the suspension of DDH-USS in the territory.

This study represents a preliminary report of late DDH diagnosis during lockdown. A comprehensive outcome evaluation of the consequences of late DDH treatment is not possible yet; a mid-term follow-up of the cases is needed and will be issue for a future paper. Due to reduction of DDH-USS during the whole of 2020 an increasing rate of late DDH diagnosis or missed diagnosis is expected in the future.

**Conclusion**

DDH-USS was the only screening in newborns which was halted during lockdown for COVID-19 in Italy. Few centres, which still performed diagnosis and treatment, were overloaded causing a delay in DDH management. In two paediatric orthopaedic centres located among the most critical areas in the Western world, late DDH diagnosis in 28-patients, with an average age at diagnosis more than three months, are reported. Due to reduction of 22% of DDH-USS during the year 2020 an increasing rate of late DDH diagnosis is expected in the future. As the duration of a lockdown is barely predictable, alternative strategies are possible to minimize the risks of both contamination and late diagnosis: 1) performing DDH-USS in a dedicated outpatient service; 2) performing DDH-USS as inpatient neonatal screening in neonatal unit care before discharge; 3) organizing a combination of the two previous strategies avoiding patient centralization in hospitals.

As showed with the present data, the balance between risks and benefits should be carefully considered in every nation, according to the local health system and screening programmes.

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**COMPLIANCE WITH ETHICAL STANDARDS**

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**OA LICENCE TEXT**

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**ETHICAL STATEMENT**

**Ethical approval:** This study was performed in accordance with the institutional review board of both centres (T and H), data were collected from registries and charts from both institutions in aggregated and anonymous form. All procedures were performed in accordance with the ethical standards of the Institutional Committee and the tenets of the 1964 Helsinki Declaration and its later amendments. **Informed consent:** Informed consent was obtained from patients’ parents/guardians.

**ICMJE CONFLICT OF INTEREST STATEMENT**

The authors declare that they have no conflict of interest. No funding was provided for the present study.

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**AUTHOR CONTRIBUTIONS**

NG: Study design, Data acquisition and analysis, interpretation, Drafting and critical revision of the text.

MDP: Study design, Data acquisition and analysis, interpretation, Drafting and critical revision of the text.
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